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Swimming and the physical, social and emotional well-being of youth with cerebral palsy

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Abstract

Cerebral palsy is the most common motor disability in childhood. The disorders of movement and posture are characterised by abnormal patterns of movement related to defective movement coordination and regulation of muscle tone. Secondary problems that may develop include perceptions of fatigue and pain. Difficulties with body function and structure affect the levels of activity and participation of the person, such as functional independence, walking ability and participation in leisure activities. These multiple facets of functioning contribute to the known physical inactivity problem apparent in youth with cerebral palsy. In addition, these low physical activity levels contribute to further deterioration of functioning. Hence, a vicious cycle of deconditioning exists. A community-based swimming programme was proposed to combat this vicious cycle. A systematic review on the effect of an aquatic intervention on the multiple facets of functioning of youth with cerebral palsy revealed no studies that measured the effect on pain, coordination and quality of life. Moreover, none of the authors reported the perceived enjoyment of the participants during the intervention. Furthermore, there was a lack of controlled studies reporting the effect of a swimming intervention on walking ability, fatigue, functional independence, self-perception, participation in leisure activities and aquatic skills in ambulatory youth with cerebral palsy. Consequently, the aim of this thesis was to investigate the effect of swimming on these multiple facets of functioning that are associated with the low physical activity levels, in youth with cerebral palsy. A randomised controlled cross-over design was implemented to investigate the effect of a 10-week swimming intervention on pain intensity, fatigue, walking ability, bilateral and upper limb coordination, functional independence, perceived competence, global self-worth, quality of life and swimming skills.

A pre-test – post-test design was used to assess the effect of taking part in a swimming programme on participation in leisure activities. Fourteen 7 to 17 year-old youth with cerebral palsy with the ability to walk with or without walking aids completed the tests on all measurement occasions. All youth had a high adherence towards the programme, participated in the intervention with high levels of enjoyment, and most youth continued to participate in swimming after completing the programme. Moreover, no adverse events due to the programme were reported and no participants withdrew from the intervention. Swimming skills improved significantly over the 10-week swimming programme, and improved significantly more than over the control period. The changes were retained throughout a 20-week follow-up period. Positive trends of improvement were evident for walking distance at maximum walking speed, upper limb coordination, functional independence in social functioning and mobility, and perceived motor competence. The intervention was not associated with increases of pain and fatigue. Participation in the swimming programme did not affect bilateral coordination, functional independence in self-care, self-perception and quality of life. The evidence of the pre-test – post-test study suggests that learning the skill of swimming encouraged participation in activities of the formal domain, active-physical and skill-based activities, and facilitated youth to engage in aquatic activities. No control data were obtained in the pre-test – post-test study; however, the results are promising in view of the known deterioration in participation with increasing age. Additionally, the present study showed that the perceptions of youth with cerebral palsy that learning a new skill is too time consuming, and that physical activity is not fun and carries a risk of injury, pain and fatigue, were eliminated in the swimming program. The consolidation of swimming skills and high levels of enjoyment during the programme, are expected to improve participation, engagement and adherence to physical activity, which was confirmed in the present study as youth participated more in aquatic activities after one year than before the start of the study. It was concluded that swimming is an enjoyable and safe community-based physical activity that may have a positive effect on the physical, social and emotional well-being in 7 to 17 year-old youth with cerebral palsy with the ability to walk. The elimination of some of the barriers confronted by youth with cerebral palsy to engaging in physical activity is important with regard to the sustainment of a physically active lifestyle. The findings suggest that participation in swimming may aid in breaking the cycle of deconditioning.

Declaration

This is to certify that that the work contained within has been composed by me and is entirely my own work. Chapter 2 of this work is based on a jointly-authored publication, which is added in the appendices of this thesis. No part of this thesis has been submitted for any other degree or professional qualification.

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A challenge, a step in the unknown, a learning process; I have enjoyed every splash.

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Chapter 1: Introduction

This chapter introduces the rationale for this thesis by discussing the multiple facets of functioning in youth with cerebral palsy (CP), as well as the framework used to organise these aspects. Additionally, the problem of physical inactivity is explained, as well as the interaction of all these factors. Finally, a solution to this problem is proposed and the purpose of study as well as the outline of the thesis are presented.

1.1 Cerebral palsy, a multifaceted disability

CP is the most common motor disability in childhood and is associated with lifelong motor impairment (Aisen et al., 2011). Rosenbaum et al. (2007) proposed the following definition: "*Cerebral palsy describes a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of cerebral palsy are often accompanied by disturbances of sensation, perception, cognition, communication, and behaviour, by epilepsy, and by secondary musculoskeletal problems*". The prevalence of CP in Europe is 1 to 2 per 1000 live births, with higher prevalence rates when birth weights are lower than 2500 g (Sellier et al., 2010; Surveillance of Cerebral Palsy in Europe, 2002), and when gestational age is lower than 36 weeks (Himpens, Van den Broeck, Oostra, Calders, & Vanhaesebrouck, 2008; Oskoui, Coutinho, Dykeman, Jetté, & Pringsheim, 2013).

Rosenbaum et al. (2007) clarified the definition by stating that abnormal gross and fine motor functioning and organisation, reflecting abnormal motor control, are the core features of CP. Loss of selective motor control, abnormal muscle tone, imbalance of power between muscle agonists and antagonists, and impaired body balance mechanisms influence the growth of the child's muscles and bones, which might result in reduced muscle elasticity, reduced joint range of motion, and disturbed bone and joint development (Koop, 2009). These motor problems can lead to difficulties with walking, feeding and swallowing, coordinated eye movements, articulation of speech, and secondary problems with musculoskeletal function, behaviour, and participation in society (Rosenbaum et al., 2007). The disorders present in CP are permanent and caused by non-progressive disturbances. This means that the disturbance is a single event or series of events, which is/are no longer active at the time of diagnosis, but produced a disruption of the normal brain structure and function that may be associated with changing manifestations over time when developing (Rosenbaum et al., 2007).

CP is a heterogeneous condition in terms of aetiology, types and severity of impairments (Rosenbaum et al., 2007), therefore, classification is necessary. The CP classification, explained by Rosenbaum, has various components, namely: motor abnormalities, accompanying impairments, anatomical and neuroimaging findings, and causation and timing. However, the classification based on neuroimaging or by cause and time of the disturbance is not yet possible due to insufficient information to provide a clear-cut classification. Traditionally, individuals with CP are classified by the dominant type of tone or movement abnormality as having a ‘spastic’, ‘dyskinetic’ or ‘ataxic’ type of CP. Additionally, the pattern and extent of the motor disorder with respect to different anatomical areas, such as unilateral or bilateral involvement, are specified. Furthermore, the functional motor abilities of the upper and lower limb in youth with CP are widely classified using the Manual Ability Classification System (MACS) and the Gross Motor Function Classification System (GMFCS) respectively (Rosenbaum et al., 2007). The GMFCS describes the gross motor functional ability of youth with CP in one of five ordered levels, where level I indicates the ability to walk without restrictions, though having limitations in more advanced gross motor skills, and level V indicates a severely limited self-mobility even with the use of assistive technology (Palisano et al., 1997). The MACS describes how children with CP use their hands to handle objects in daily activities in five levels: level I indicates the ability to handle objects easily and successfully and level V describes children that do not handle objects and have severely limited ability to perform even simple actions (Eliasson et al., 2006). Classification of youth with CP also includes the presence or absence of accompanying impairments (e.g. visual impairments) (Rosenbaum et al., 2007).

Aisen et al. (2011) described the rehabilitative management of the abnormal gross and fine motor control and secondary difficulties in youth with CP. The management includes: physical therapy focussing on gross motor skills, muscle strengthening and the provision of mobility devices and other equipment needs, occupational therapy focussing on upper-extremity functions such as feeding and dressing, and speech and language therapy addressing articulation, readiness for school, etc. Stretching and flexibility exercises remain integral components of most physical therapy programmes, despite inconclusive evidence for their effectiveness. Regular exercise is necessary for the health of children and adults, but in those with CP the ability to exercise is adversely affected by their motor impairment (Aisen et al., 2011). Damiano (2006) recommended implementing physical activity into the rehabilitation programme. Other approaches include medication and orthopaedic surgical interventions to affect muscle tone and to alleviate severe mechanical impairments respectively (Aisen et al., 2011).

The following sections elaborate on the multiple facets of functioning of youth with CP, using the international classification of functioning, disability and health framework of the World Health Organization (2007).

1.1.1 The international classification of functioning, disability and health

As Rosenbaum et al. (2007) have defined, CP incorporates a group of motor disorders that cause activity limitations and are often accompanied by additional disturbances. In this thesis the ‘International Classification of Functioning (ICF), Disability and Health – Children and Youth Version’ is used as a framework to organise the group of disorders and multiple facets of functioning in children and adolescents with CP, in a meaningful and interrelated way (Figure 1.1) (World Health Organization, 2007).

The World Health Organization (2007) described the ICF as consisting of two parts: one part covers components of functioning and disability and one part covers the contextual factors. The functioning and disability part consists of a ‘body’ component and an ‘activities and participation’ component. More specifically, the ‘body’ component comprises ‘body functions’, which refers to the physiological functioning of the body, and ‘body structures’, which refers to the anatomical parts of the body. Activities are the execution of a task or action and participation is the involvement in life situations. As it is difficult to distinguish between the activities and participation domains, the 'activities and participation' component covers a full range of life areas in a single list. The second part, covering the contextual factors, incorporates the environmental and personal factors. Environmental factors are the physical, social and attitudinal contexts (external to the individual) in which an individual conducts his/her life, and personal factors include the particular background of an individual's life and living, and features of the individual that are not part of a health condition (gender, race, age, lifestyle, habits, etc.) (World Health Organization, 2007).

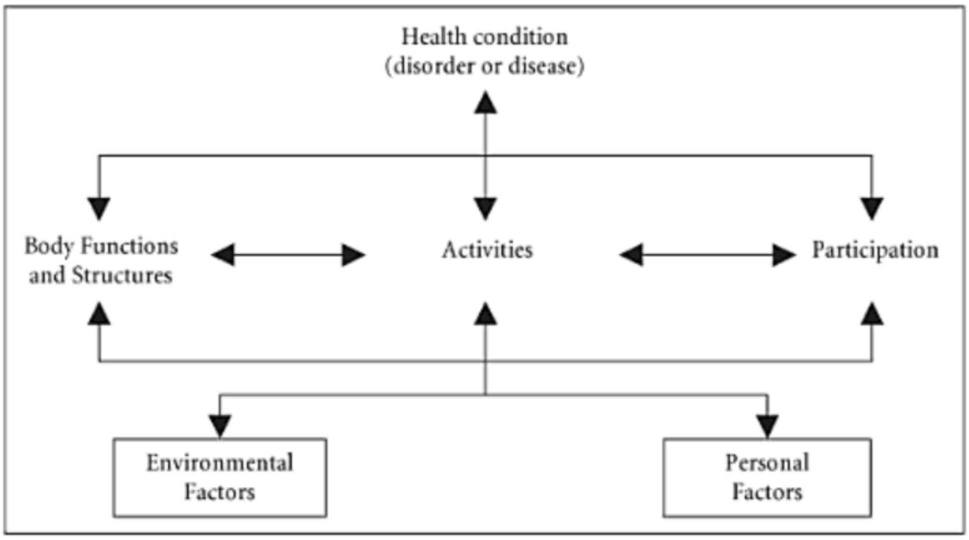


Figure 1.1 The international classification of functioning, disability and health (World Health Organization, 2007).

The following paragraphs expand on the issues that youth with CP encounter, and categorise these issues within the ICF components.

The disorders of movement and posture present in youth with CP are characterised by abnormal movement patterns related to defective coordination and/or regulation of muscle tone (Bax et al., 2005) that are categorised under the 'body' component of the ICF framework. A cascade of muscle and bone consequences follows the altered brain function in CP. Loss of elasticity in combination with altered spontaneous limb movement reduces muscle length and limits joint motion, leading to contractures. The bones to which the muscles are attached change their growth: infantile bone shapes fail to resolve and some bones develop torsions (Koop, 2009). Altered bone shapes change the effect of muscles, leading to lever arm dysfunction, which reduces the ability to generate an effective moment. Stiff soft tissues including muscles and bony and joint abnormalities impair the biomechanics of gait (Levitt, 2010). Human walking is a complex interaction between the central nervous system and the peripheral musculoskeletal system and can be categorised under the 'activities and participation' component of the ICF framework. Seventy per cent of the European youth with CP are able to walk with or without aids (Beckung, Hagberg, Uldall, & Cans, 2008), but a lower walking speed than the typically developing youth is apparent (Abel & Damiano, 1996). This lower walking speed might be due to the inappropriate muscle activation that increases the use of energy resources (Koop, 2009). Production of functional gait requires sufficient energy, the ability to balance, central program generators to provide motor control, and stable mechanical structures to support the force output (Miller, 2005). Additionally, 45% of the Norwegian adults with CP reported a deterioration of walking skills, with an onset of deterioration between 15 and 34 years of age for 64% of these adults (Jahnsen, Villien, Egeland, Stanghelle, & Holm, 2004).

A factor that matters most to a child or adolescent and their family is the ability to perform daily activities (Levitt, 2010), also known as functional independence, which can be categorised under the 'activities and participation' component of the ICF framework. Mesterman et al. (2010) reported that 30% to 50% of the Israeli 8 to 30 year-olds with CP require assistance in activities of daily living. For the tasks 'dressing', 'shower' and 'mobility outside of home' 50% to 60% of the participants aged below 18 years reported the need for assistance (Mesterman et al., 2010). After achieving a maximum independence level, social function and mobility skills have been reported to deteriorate in youth with CP from the age of 14 years onwards (Kerr, McDowell, Parkes, Stevenson, & Cosgrove, 2011). Nieuwenhuijsen, Donkervoort, Nieuwstraten, Stam, and Roebroeck (2009) reported that 70% of Dutch 18 to 22 year-olds with CP experience problems with activities in daily life.

Secondary problems that develop mainly in late childhood include fatigue and pain (Levitt, 2010), which can be categorised under the 'body' component of the ICF framework. Pain has been reported to be present in 60% of the European 8 to 12 year-old children with CP (Parkinson, Gibson, Dickinson, & Colver, 2010) and in 74% of the European 13 to 17 year-old adolescents with CP (Parkinson et al., 2013). Causes of pain in ambulatory youth with CP have been reported by Canadian physicians to be focal muscle spasms, muscle weakness, overuse and fatigue, and abnormal gait patterns (Penner, Xie, Binepal, Switzer, & Fehlings, 2013). Fatigue can be a result of the high energy requirement of ambulatory youth with CP (Dallmeijer & Brehm, 2011; Fowler et al., 2007) to maintain ambulatory capacity (Bottos & Gericke, 2003). Following the proposal of Kluger, Krupp, and Enoka (2013) concerning the taxonomy of fatigue, there are two large distinctions in the definition of fatigue, being the perception of fatigue and the performance fatigability. The former refers to the subjective increasing sense of effort, sensations of weariness, or exhaustion. Fatigability is defined as "*the magnitude or rate of change in a performance criterion relative to a reference value over a given time of task performance*" (Kluger et al., 2013). Thirty per cent of 406 Norwegian adults with CP reported substantial perceptions of fatigue, and for the entire sample the reported physical fatigue was significantly higher than for the general Norwegian population (Jahnsen, Villien, Stanghelle, & Holm, 2003). Similarly, Dutch adults with CP reported significantly more chronic pain and higher perceptions of fatigue than reference samples (Van der Slot et al., 2012). These factors may affect the energy a person has to participate in therapy programmes, and they have to be taken into account when designing such programmes (Levitt, 2010). According to adolescents and young adults with CP the physical therapy programme during childhood induces perceptions of fatigue and fatigability and is the cause of pain and physical distress (Jahnsen, Villien, Aamodt, Stanghelle, & Holm, 2003; Redmond & Parrish, 2008).

In addition to the components of functioning and disability, it is important to address the contextual factors. Self-concept, referring to an overarching view of the self, and self-esteem, reflecting a person's evaluative assessment of themselves (Butler & Gasson, 2005), have been reported being lower in youth with CP than in their age-matched peers (Riad, Brostrom, & Langius-Eklof, 2013; Russo, Goodwin, et al., 2008; Soyupek, Aktepe, Savas, & Askin, 2010), and are categorised under the personal contextual factors of the ICF framework. Specifically, perceived scholastic competence, athletic competence, social acceptance and physical appearance have been reported being lower in youth with CP than in their age-matched peers (Russo, Goodwin, et al., 2008; Shields, Loy, Murdoch, Taylor, & Dodd, 2007).

Body functions and structures, activities, personal and environmental factors all affect the involvement in life situations, better known as participation (World Health Organization, 2007). Participation is fundamentally important to a child's development, and without adequate opportunities to participate "*people are unable to explore their social, intellectual, emotional, communicative, and physical potential and are less able to grow as individuals*" (King et al., 2003). There is a body of evidence showing that participation in leisure activities of youth with CP is less varied and less intense than their peers, and happens with family close to home rather than with friends in the broader community (Bult et al., 2010; Engel-Yeger, Jarus, Anaby, & Law, 2009; Imms, Reilly, Carlin, & Dodd, 2008). Participation of youth with CP in formal activities (structured, involving rules or goals) has been described as less diverse and less intense than in informal activities (spontaneous, involving little planning, initiated by the child) (Imms et al., 2008; Law et al., 2006; Majnemer et al., 2008). Participation diversity and intensity is known to be particularly low in skill-based and active-physical activities, although youth with CP report high levels of enjoyment for participation in a wide variety of activity types (Imms et al., 2008; Law et al., 2006; Majnemer et al., 2008).

One composite variable that has not been classified within the ICF framework to date is quality of life (QoL). It is defined as a "*composite emergent and changing phenomenon of all aspects of functioning that may be impacted by a person's health condition, personal and environmental factors*" and has been proposed to be included in the ICF framework (McDougall, Wright, & Rosenbaum, 2010). Health-related QoL has been reported, by both the children and adolescents themselves and their parents, to be lower for youth with CP than for youth without a disability (Maher, Olds, Williams, & Lane, 2008; Russo, Goodwin, et al., 2008; Soyupek et al., 2010; Vargus-Adams, 2005; Varni et al., 2005), in particular for the aspects of physical, emotional, social and school functioning (Du, McGrath, Yiu, & King, 2010).

In summary, CP is the most common motor disability in childhood and is characterised by a complex group of disorders and multiple facets of functioning that interfere with physical, emotional and social well-being. Using the ICF framework: diverse impairments of body function and structure (pain, perceptions of fatigue and coordination) in addition to activity limitations and participation restrictions (walking speed, functional independence and participation in leisure activities) have been identified in youth with CP. Additionally, the contextual personal factors self-concept and self-esteem, as well as QoL, have been reported being lower than in age-matched peers (Russo, Goodwin, et al., 2008; Soyupek et al., 2010). The environmental contextual factors, which are external to the individual, were not included in the scope of this thesis.

Furthermore, the ICF emphasizes the interrelation and dynamic interaction between the components of its framework (Figure 1.1). These interactions are discussed in the following sections.

1.1.2 Interaction of the various components of the framework

The ICF framework reflects that within a context, a health condition originates from the interaction between body functions and structures, and activities and participation (World Health Organization, 2007). It proposes that an individual's functioning is an interaction or complex relationship between the health condition and the contextual factors. The interaction among these entities is dynamic, as interventions in one entity have the potential to modify one or more of the other entities. The potential interactions of the multiple facets of functioning in youth with CP found in scientific research are discussed subsequently and are displayed in Figure 1.2.

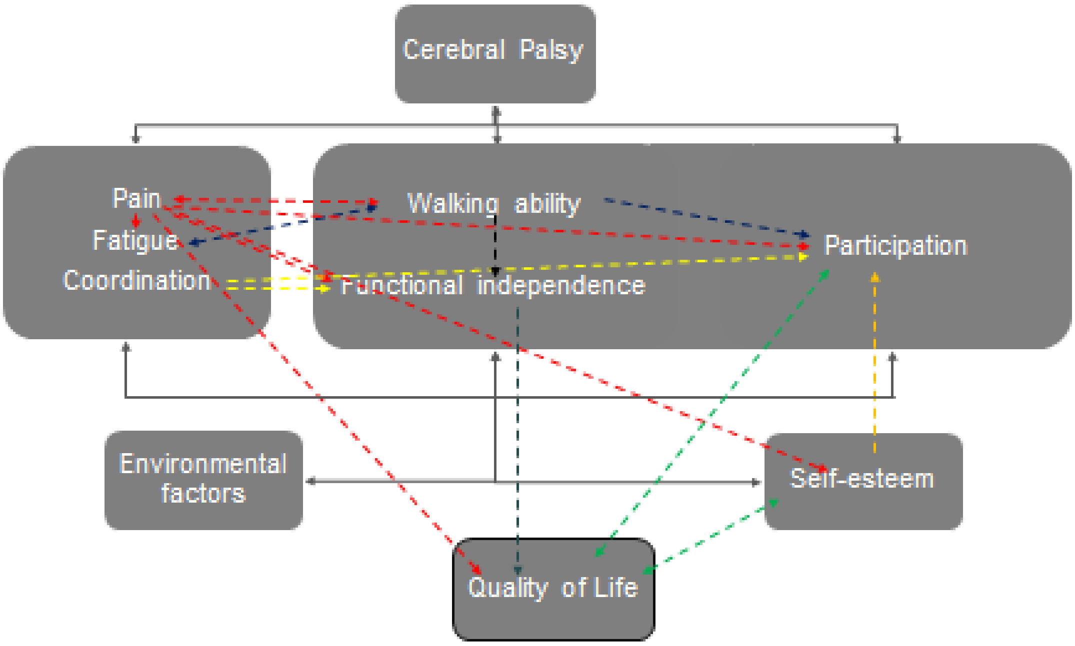


Figure 1.2 The multiple facets of functioning are displayed according to the 'International Classification of Functioning (ICF), Disability and Health' framework in addition to quality of life. The interactions of these components are displayed with arrows, using different colours for clarity.

Manual ability and gross motor function levels predict 66% of the variance in functional independence in self-care and 76% of the functional independence in mobility in Swedish youth with CP, respectively (Öhrvall, Eliasson, Löwing, Ödman, & Krumlinde-Sundholm, 2010). Consequently, functional independence and coordination need to be considered together. Musculoskeletal pain in youth with CP has been reported being intensified by walking or running (Ramstad, Jahnsen, Skjeldal, & Diseth, 2011). Canadian physicians reported muscle fatigue and abnormal gait patterns as causes of pain present in youth with CP (Penner et al., 2013). Research in adults with CP shows that pain, in turn, contributes to locomotion deterioration (Morgan & McGinley, 2013), and that it predicts walking ability (Maanum, Jahnsen, Froslie, Larsen, & Keller, 2010) and perceived physical fatigue (Jahnsen, Villien, Stanghelle, et al., 2003). Furthermore, a higher energy cost during walking contributes to fatigability and perceptions of fatigue in youth with CP (Dallmeijer & Brehm, 2011; Fowler et al., 2007), while fatigability and perceptions of fatigue predict ambulatory capacity and locomotion deterioration in adulthood (Bottos & Gericke, 2003; Morgan & McGinley, 2013). It can be concluded that pain, perceptions of fatigue and walking ability are strongly interrelated. Soyupek et al. (2010) reported that QoL and self-concept were correlated in a sample of Turkish youth with CP aged 8 to 19 years.

Furthermore, pain has been found to influence participation (Fauconnier et al., 2009; Ramstad, Jahnsen, Skjeldal, & Diseth, 2012), functional independence (Tarsuslu & Livanelioglu, 2010), self-concept and QoL (Russo, Miller, Haan, Cameron, & Crotty, 2008). Youth with a mobility impairment who are more dependent in activities of daily living have been shown to report a significantly lower score on the QoL subscale 'psychological well-being' (Jemta, Fugl-Meyer, Oberg, & Dahl, 2009). According to Palisano et al. (2009) reduced walking speed, endurance, and economy can limit the ability of children and adolescents with GMFCS levels II and III to keep up with peers, especially outdoors and in the community. Additionally, participation levels have been shown to be influenced by motor proficiency (in typically developing children) (Okely, 1999), physical functioning and sports competence (in adolescents with CP) (Kang et al., 2010), movement ability (in children with CP) (Bult et al., 2013) and perceived athletic and scholastic competence of children and adolescents with disabilities (Barnett, Morgan, van Beurden, & Beard, 2008; King et al., 2003). Finally, Groff, Lundberg, and Zabriskie (2009) reported that participation in adapted sports is associated with QoL.

As the ICF framework suggests, and evidence from research supports, the multiple aspects of the physical, social and emotional well-being of youth with CP interact.

In summary, the multifaceted problems apparent in youth with CP were discussed using the framework of the ICF. Most children and adolescents with CP are functionally dependent (Mesterman et al., 2010), which partly can be explained by the abnormal motor control that is the core characteristic of CP (Öhrvall et al., 2010; Rosenbaum et al., 2007). Additionally, children and adolescents with CP have been reported having substantial levels of pain and fatigue (Bottos & Gericke, 2003; Parkinson et al., 2013; Parkinson et al., 2010), and a lower walking speed than typically developing children (Abel & Damiano, 1996). All three factors: perceptions of fatigue, pain and walking ability are known to be interrelated (Dallmeijer & Brehm, 2011; Jahnsen, Villien, Stanghelle, et al., 2003; Morgan & McGinley, 2013; Ramstad et al., 2011). Also, self-concept and QoL have been reported being interrelated and being lower in youth with CP than in typically developing youth (Soyupek et al., 2010). Finally, youth with CP participate in leisure activities with less frequency and less variety than typically developing peers, and with family close to home rather than with friends in the community (Imms et al., 2008).

The following section introduces the problem of physical inactivity and its relevance to youth with CP and their complex set of issues. Physical inactivity is a global public health problem and an important risk factor for multiple causes of death and chronic morbidity and disability (Bull et al., 2004).

1.2 A vicious cycle of deconditioning

Children and adolescents with CP are considerably less active (Bjornson, Belza, Kartin, Logsdon, & McLaughlin, 2007; Carlon, Taylor, Dodd, & Shields, 2013) and have lower physical fitness levels (Rimmer, 2001; Verschuren & Takken, 2010) than their able-bodied peers. This physical inactivity and low physical fitness can result in a decline in health and physical function, e.g. deterioration of walking skills (Bottos & Gericke, 2003; Jahnsen et al., 2004), and further inactivity. This vicious cycle represents a cycle of deconditioning (Durstine et al., 2000). As the level of physical activity in childhood predicts the activity level in adulthood (Telama et al., 2005), this inactivity involves numerous health risks. Perceived barriers reported by youth with CP to engaging in physical activity include pain during exercise, fear of increased risk of an injury, beliefs that learning a motor skill is too time-consuming and the perception of physical activity and sports as not being fun (Verschuren, Wiart, Hermans, & Ketelaar, 2012). As the cycle of deconditioning implies, the multiple facets of functioning that characterise the disability are influencing and are influenced by the physical activity and physical fitness levels (Figure 1.3). The following section expands the role of the physical, emotional and social factors that were introduced in section 1.1, in the vicious cycle of deconditioning.

1.2.1 Influencing factors

A lack of physical activity has been found to be associated with perceived physical fatigue and to contribute to locomotion deterioration in Norwegian adults with CP (Jahnsen et al., 2004; Jahnsen, Villien, Stanghelle, et al., 2003). Additionally, Maltais, Dumas, Boucher, and Richards (2010) reported that an increase in pain and perceptions of fatigue is associated with a higher chance of inactivity among Canadian adult walkers with CP. Verschuren et al. (2012) listed the barriers and facilitators of Dutch youth with CP to engage in physical activity, of which the personal barriers included lack of energy/fatigue, pain and feeling insecure or being ashamed. Furthermore, a study of Turkish adolescents with CP by Tarsuslu and Livanelioglu (2010) indicated that the more dependent in locomotion, mobility and self-care, the lower the physical activity levels. Therefore it is important to address pain and fatigue, as well as limitations in walking ability and functional independence, as these factors are associated with physical activity levels in youth and adults with CP.

The level of motor coordination in Australian extremely-low-birth-weight children has been found to predict fitness levels (Burns et al., 2009). In the typically developing North American population, the level of motor coordination has been found to have a positive association with physical activity (Wrotniak, Epstein, Dorn, Jones, & Kondilis, 2006). Additionally, higher competence in activities and sports has been shown to enable North American youth with CP to engage in a greater variety of activities with friends (Kang et al., 2010). Perceived sports competence has been reported acting as a mediator between childhood motor skill proficiency, and adolescent physical activity and fitness levels in the general Australian population (Barnett et al., 2008). Therefore, motor coordination as well as perceived and actual sports competence are important factors in addressing physical activity, physical fitness and participation levels. Participation in a sport club has been reported to have a significant association with ambulatory activity in the weekend in Dutch youth with CP (Van Wely, Becher, Balemans, & Dallmeijer, 2012).

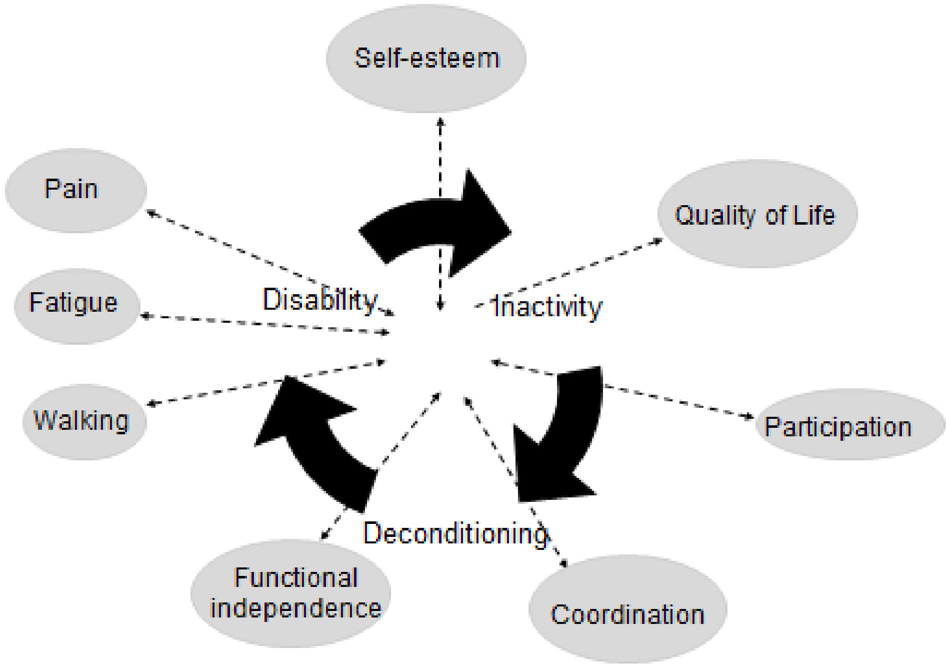


Figure 1.3 Multiple facets of functioning associated with the cycle of deconditioning.

Higher physical fitness levels have been found to be associated with higher values of self-esteem and body satisfaction (Greenleaf, Petrie, & Martin, 2010). Physical activity has been found to be a strong predictor in the prevention of chronic pain, fatigue and deterioration of locomotor skills in Norwegian adults with CP (Jahnsen, Villien, Aamodt, et al., 2003). This implies that aiming to improve physical activity and physical fitness levels will contribute to combatting a wider set of problems, implicating a reversal of the vicious cycle of deconditioning.

In summary, youth with CP are insufficiently physically active and fit (Bjornson et al., 2007; Carlon et al., 2013; Rimmer, 2001; Verschuren & Takken, 2010). The complex set of problems in youth with CP contributes to this physical inactivity, and low physical fitness and participation levels, and vice versa. Hence, a vicious cycle of deconditioning exists in youth with CP.

The following section discusses the implementation of community-based exercise and sport programmes into the rehabilitation programme with the aim of breaking and reversing this cycle of deconditioning (Bania, Dodd, & Taylor, 2011; Fowler et al., 2007; Rimmer, 1999, 2005).

1.3 Swimming to break the cycle of deconditioning

Previous sections 1.1 and 1.2 discussed the complex set of problems that youth with CP encounter, and the cycle of deconditioning that is present in youth with CP. The following section introduces the implementation of community-based exercise and sport programmes to address the components of this cycle of deconditioning (Bania et al., 2011; Fowler et al., 2007; Rimmer, 1999, 2005).

A systematic review of Verschuren, Ketelaar, Takken, Helders, and Gorter (2008) on exercise in youth with CP reported 20 research trials (of which six were randomised controlled trials) that assessed the effect of exercise on body function and activities. The described exercise programmes involved strength training, aerobic training, and mixed programmes (aimed to improve one or more components of physical fitness such as muscle strength, aerobic and anaerobic capacity, flexibility, and body composition). The aerobic training programmes included cycling, running, swimming and treadmill training. The review reported positive effects on aerobic capacity, gross motor function, muscle strength, muscle tone, stride length and self-perception in physical appearance, scholastic competence and social acceptance. Additionally, a positive effect on perceived athletic competence has been reported after 8 months of aerobic and anaerobic exercises in children and adolescents with CP (Verschuren et al., 2007), and on QoL after a 12-week stationary cycling intervention in youth with CP (Demuth, Knutson, & Fowler, 2012). A systematic review by Ekeland, Heian, and Hagen (2005) reported a positive effect of exercise (aerobic, strength training, skills training, and combinations of these) on self-esteem in typically developing children and adolescents. More generally, participation in exercise has been found to promote physical, emotional and social well-being of youth with disabilities (Murphy & Carbone, 2008).

Due to the paucity of accessible programmes and other significant barriers (e.g. lack of motivation, time, or information) (Buffart, Westendorp, van den Berg-Emons, Stam, & Roebroeck, 2009; Verschuren et al., 2012), participation in exercise programmes needs to be facilitated in youth with CP. Additionally, sustaining a physically active lifestyle is essential (in youth with CP) to achieve and maintain functional capability (Riner & Sellhorst, 2013). Motivation and enjoyment are facilitators for engaging in physical activity, for adhering to physical activity and for continuing committing to sport (Buffart et al., 2009; Martin, 2006; Redmond & Parrish, 2008).

A recent review of Riner and Sellhorst (2013) recommended physical activity and exercise programmes for youth with CP to be enjoyable, to be within the child's capabilities, and to include only activities with limited risk of falling or injury. However, the levels of enjoyment or satisfaction of the assessed intervention programmes have not been reported in previous studies, reported in the extant literature such as the systematic reviews of Verschuren et al. (2008) and Ekeland et al. (2005).

Facilitators for engaging in physical activity include '*having fun*', '*different from therapy*', '*learning a skill*' and '*independence and freedom*' (Shimmell, Gorter, Jackson, Wright, & Galuppi, 2013). Swimming is a community-based exercise incorporating all of these characteristics. It is believed to be fun, not to increase pain during exercise and not to increase the risk for injury (Retarekar, Fragala-Pinkham, & Townsend, 2009). Moreover, swimming can be introduced at a young age and offers recreational opportunities that can encourage sustaining a physically active lifestyle.

Water can facilitate movement due to its physical and fluid dynamic properties; Buoyancy frees a body submerged in water from the downward pull of gravity, resulting in decreased joint loading (Bates & Hanson, 1996; Irion, 2009), weight relief and ease of movement (Cole & Becker, 2004), reflecting a decrease in biomechanical constraints. This allows youth with CP to exercise in water with more freedom than on land (Kelly & Darrah, 2005), without increasing the risk of falling. Hydrostatic pressure, a second physical property of water, can provide enhanced tactile input (Irion, 2009). Therefore, water can offer feedback about body sensation, body awareness, joint positions, spatial awareness and postural control (Gjesing, 2000) whilst learning the rhythmic inter-coordination of upper and lower limbs required for the skill of swimming. Moreover, swimming, involving skill-learning, enhances motor proficiency. The latter has been reported to have a positive association with levels of fitness (Burns et al., 2009), activity (Wrotniak et al., 2006) and participation (Kang et al., 2010; Okely, 1999), and this association is positively mediated by perceived sports competence (Barnett et al., 2008). Perceived sports competence is expected to be high, since water is an environment where everybody is more equal than on land. Finally, learning how to swim allows a person to engage in other physical activities performed in the water, such as sailing, kayaking, surfing, rowing, scuba diving, playing water polo, etc.

Three systematic reviews summarising the effects of aquatic interventions in youth with neuromotor impairments (Getz, Hutzler, & Vermeer, 2006) and CP (Gorter & Currie, 2011; Jorgić, Dimitrijević, Lambeck, et al., 2012) reported significant improvements in aquatic skills, muscle strength, gross motor function, walking capacity, vital capacity, self-esteem and functional independence after various types of aquatic interventions, including aerobic training such as length swimming, water walking/running, relay races; anaerobic activities such as jumping; and strength or resistance training in the water. Although statistical significance was not reached, participation in physical activity improved after the interventions. The literature provides preliminary evidence of the effectiveness of various aquatic interventions on body functions and activity limitations. Nevertheless, in most of the studies case series designs, including small samples, have been implemented, and several relevant outcome measures such as pain, coordination and QoL have not been assessed.

In summary, exercise is a possible solution to counteract the deconditioning associated with cerebral palsy. However, engagement in physical activity needs to be facilitated and adherence to a physically active lifestyle is essential. The proposed physical activity in the present study is swimming, as it can be presented as enjoyable exercise without increasing pain during exercise or increasing the risk of injury, and therefore eliminates some of the barriers to engaging in physical activity perceived by youth with CP (Verschuren et al., 2012). Skill-learning and the levels of enjoyment influence participation (Kang et al., 2010; Okely, 1999), adherence (Redmond & Parrish, 2008), and the commitment to sport (Martin, 2006). Additionally, swimming and other aquatic interventions have been reported to have a positive effect on the multiple facets of functioning of youth with CP that contribute to the vicious cycle of deconditioning. However, due to a lack of controlled trials on the effects of swimming on pain, fatigue, walking ability, coordination, functional independence, self-esteem, QoL and participation in leisure activities, a conclusion is difficult to draw.

1.4 Purpose of study

Children and adolescents with CP encounter a complex set of issues that interfere with physical, emotional and social well-being. These issues are interrelated and associated with physical inactivity and the vicious cycle of deconditioning. A community-based swimming programme was proposed to combat the cycle of deconditioning as engagement and adherence are expected to be facilitated, and was proposed to have a positive effect on the multiple facets of functioning at various levels of the ICF framework. Therefore, the purpose of this study was to investigate the effect of swimming on pain, (perceptions of) fatigue, walking ability, functional independence, coordination, perceived competence and global self-worth, QoL, aquatic and swimming skills, as well as on participation in leisure activities, in youth with CP with the ability to walk. Additionally the retention of possible gains was investigated. Furthermore, the enjoyment of the swimming sessions was assessed, since motivation and enjoyment are essential to sustain a physically active lifestyle.

1.5 Outline of the thesis

Chapter 1 provided an introduction to the physical, emotional and social issues that youth with CP encounter and the proposed intervention, swimming, to break the cycle of deconditioning. Chapter 2 gives an overview of the literature on swimming and the effect on these physical, emotional and social components (pain, perceptions of fatigue, walking speed, coordination, functional independence, self-perception, QoL and participation), as well as on swimming skills, and the enjoyment of the programmes. The specific research questions are formulated following the conclusions drawn from the review. Chapters 3 to 10 describe the methods and results of the research study that was conducted to investigate the research questions. The methods of the research study, the included population, the intervention used and the assessed outcomes are explained in Chapter 3. The subsequent chapter, Chapter 4, describes the results of the present study related to the characteristics of the participants and the compliance with the programme. Chapters 5 to 10 report and discuss the results related to each research question. A general discussion of the thesis is provided in Chapter 11, followed by the conclusion in Chapter 12.

Chapter 2: Literature review and research questions

The previous chapter described the complex set of problems encountered by youth with cerebral palsy (CP), as well as the cycle of deconditioning and the rationale for proposing swimming as a valuable intervention programme to address these problems. This chapter reviews the literature, published since 2000, on the effects of swimming on the physical, emotional and social well-being of youth with CP. The issues were organised in Chapter 1 using the international classification of functioning, disability and health framework (World Health Organization, 2007). They are pain, perceptions of fatigue, walking ability, coordination, functional independence, self-esteem, quality of life (QoL) and participation. Additionally, this chapter reviews the literature on the progression of swimming skills and enjoyment of the aquatic interventions in youth with CP. Following the conclusions that are drawn from this review, the research questions are formulated.

The literature review addressed the general question 'can swimming be used to influence the complex set of problems associated with the vicious cycle of deconditioning that children and adolescents with CP encounter?'.

2.1 Protocol for the literature search, study selection, quality assessment & data extraction

The systematic review was conducted according to the guidelines of the University of York (Centre for Reviews and Dissemination, 2008) on undertaking reviews in health care, as suggested by NICE (National Institute for Health and Clinical Excellence, 2006).

The electronic search used the following databases: Cochrane, PEDro, Pubmed, ScienceDirect, SPORTSdiscus, Medline and CINAHL. Several search terms were used: "hydrotherapy", "aquatic therapy", "water exercise", "aquatics", "adapted aquatics", "aquatic exercise", "swimming", "Halliwick", "Watsu", "Ai chi" and "Bad Ragaz Ring Method" in combination with "cerebral palsy", "neuromotor", "neuromuscular" and "neurologic". Original research articles including children and adolescents (0 to 21 years old) with a disease of the nervous system, an aquatic or swimming intervention and publication dates between January 2000 and April 2013 were included. All types of study designs were incorporated. Research articles investigating a healthy population and policy statements without an actual intervention were excluded.

Two reviewers searched the literature independently and assessed whether the research articles met the inclusion criteria. Any discrepancies were resolved through consultation with a third reviewer. The search was limited to English, French, Dutch and Latvian language papers.

The algorithm for classifying study designs from The National Institute for Health and Clinical Excellence recommended by Scottish Intercollegiate Guidelines Network (2008) was used to identify the study designs. Each article was allocated a level of evidence (Sackett, 1989; Siebes, Wijnroks, & Vermeer, 2002): Randomised Controlled Trials (RCTs) and non-RCTs were marked as Level I and II, respectively, observational case studies with control participants as level III, and before-after studies, case series and case reports as levels IV and V, respectively. In addition, RCTs were evaluated by two independent assessors using the PEDro scale, a valid scale to measure the methodological quality of clinical trials (de Morton, 2009).

The research population and the participants' age range were extracted. The different types of aquatic interventions reported in the articles were classified in one of the following categories: 'Halliwick concept' (International Halliwick Association Education and Research Committee, 2010), 'aquatic physiotherapy', 'swimming (exercises and techniques)', 'exercises in water', 'walking activities in water', 'games/play (in water)'. The addition of songs or music, breathing exercises, stretching, relaxation, and/or exercises or games out of the pool was noted. The duration and intensity of the programmes as well as the description of the swimming pool where the intervention took place, were extracted. The outcome measures were categorised within the International Classification of Functioning (ICF), Disability and Health (World Health Organization, 2007).

2.2 Results of the literature search

The results of the literature search are presented in Figure 2.1. From the original 951 research articles, 934 were excluded after further inspection.

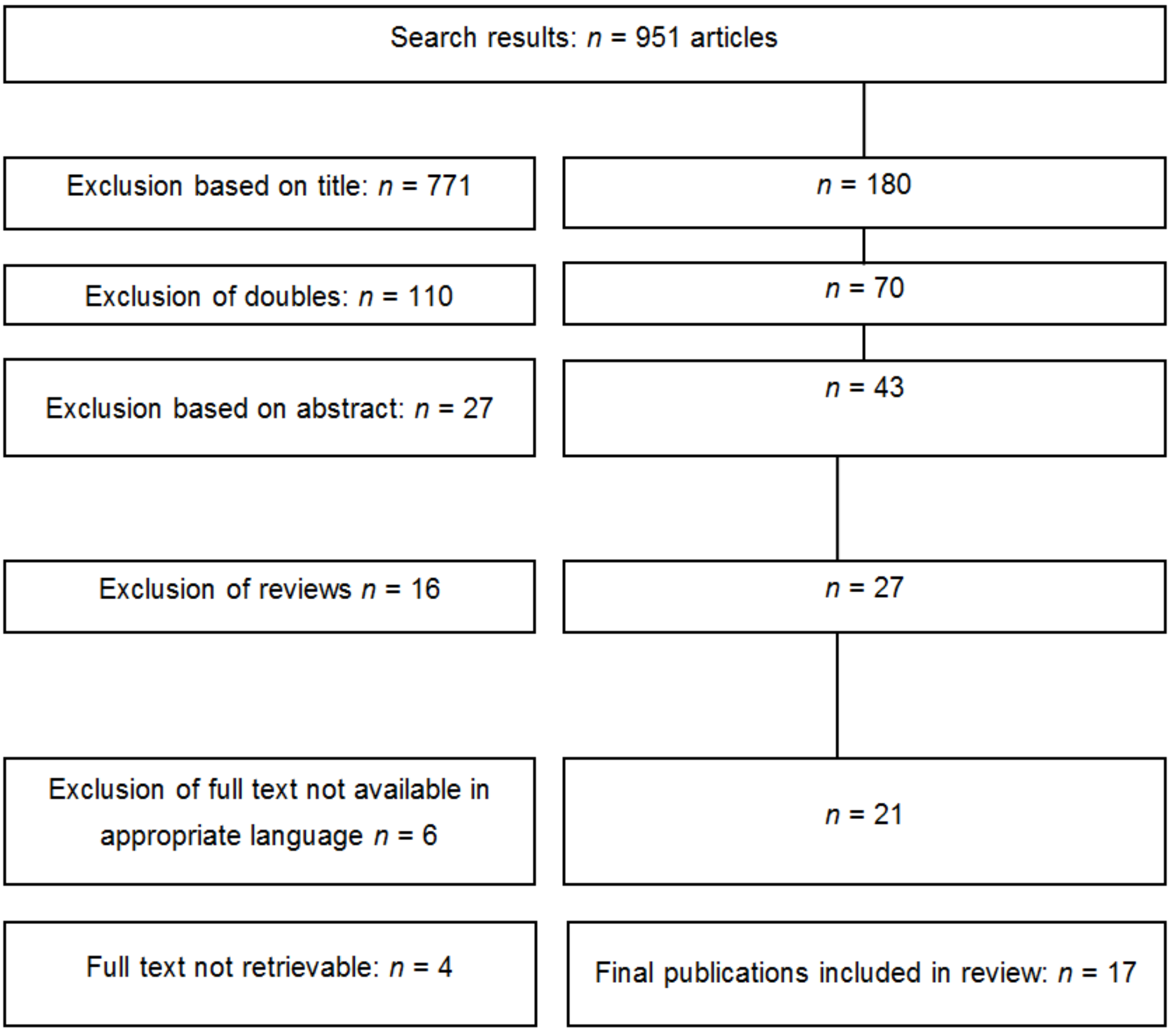


Figure 2.1 Flowchart of the literature search.

2.2.1 Study designs and quality

The number of research articles for each study design are shown in Table 2.1. Six of the 17 retrieved research articles had evidence levels I and II, which are allocated to Randomised Controlled Trials (RCTs) and non-RCTs respectively (Sackett, 1989; Siebes et al., 2002). Consequently, only 6 of the 17 intervention studies included a comparison group. Getz, Hutzler, and Vermeer (2007) and Getz, Hutzler, Vermeer, Yarom, and Unnithan (2012) compared an aquatic intervention and a land-based exercise intervention. The other four studies included a control group who did not receive any additional therapy besides the usual care (Chrysagis, Douka, Nikopoulos, Apostolopoulou, & Koutsouki, 2009; Dimitrijević et al., 2012; McManus & Kotelchuck, 2007; Őzer et al., 2007). The three RCTs (evidence level I) (Chrysagis et al., 2009; Dimitrijević et al., 2012; Őzer et al., 2007) scored 6/10, 4/10 and 5/10 on the PEDro scale, respectively. More than two thirds of the identified articles had evidence levels IV or V, implying case series and case reports, which are of a low methodological quality (Sackett, 1989; Siebes et al., 2002).

Table 2.1 The number of articles per study design.

Level of evidence	Intervention studies	*n* = 17
I	RCTs	3
II	non-RCTs	3
III, IV, V	Before-after studies, case series and case studies	11

Note. Levels of evidence according to Sackett, 1989 and Siebes, Wijnroks, & Vermeer, 2002.

2.2.2 Participant characteristics

The children and adolescents participating in the research interventions were aged from six months to 21 years. Most research samples (11 out of 17) included only youth with CP. The other samples also included youth with other neuromotor disorders (the number of these participants in between brackets), namely: spinal muscular atrophy (*n* = 2), meningomyelocele (*n* = 6), developmental delay (*n* = 12), muscular myopathy (*n* = 3), prematurity (*n* = 7), hypotonia (*n* = 2), chromosomal anomalies (*n* = 7), sensory integration deficits (*n* = 4) and Prader-Willi Syndrome (*n* = 1) in addition to youth with CP. Some samples also included youth with mental and behavioural disorders such as Autism Spectrum Disorder (*n* = 10), Down syndrome (*n* = 4), learning disabilities (*n* = 2), Pervasive Developmental Disorder Not Otherwise Specified (*n* = 3) and musculoskeletal disorders such as Juvenile Ideopatic Arthritis (*n* = 1) and oto-palatal-digital syndrome (*n* = 1). The maximum number of youth with CP participating in a research trial was 29 (in the RCT by Dimitrijević et al. (2012)).

2.2.3 Types of aquatic interventions

Many authors reported more than one type of intervention that was used during their programme (Table 2.2). Swimming was the main intervention types reported in the three RCTs (Chrysagis et al., 2009; Dimitrijević et al., 2012; Őzer et al., 2007). The ‘Halliwick concept’ was used in three publications (Getz et al., 2007; Getz et al., 2012; Jorgić, Dimitrijević, Aleksandrović, et al., 2012). In nine studies ‘exercises in water’ was the main content of the programme (Ballaz, Plamondon, & Lemay, 2011; Fragala-Pinkham, Dumas, Barlow, & Pasternak, 2009; Fragala-Pinkham, Haley, & O'Neil, 2008; Fragala-Pinkham, O'Neil, & Haley, 2010; Kelly, Darrah, Sobsey, & Legg, 2009; McManus & Kotelchuck, 2007; Retarekar et al., 2009; Salem & Gropack, 2010; Thorpe, Reilly, & Case, 2005). One article implemented ‘aquatic activities’, but programme details were not reported (Aidar et al., 2007). The detailed information for each intervention, provided by the author, is summarised in Appendix A. The duration of the programmes ranged from 6 to 36 weeks, with 1 to 5 sessions a week and 30 to 60 min per session. The type of swimming pool was described in 12 articles, while water temperature was reported in 10 articles.

Table 2.2 The intervention categories reported in the articles.

Intervention type / Author(s), Year	Halliwick concept	Aquatic physiotherapy	Swimming (exercises and techniques)	Exercises in water	Walking activities in water	Games/play activities (in water)	Exercises, games out of the pool	+ Songs or music	Breathing exercises	Stretching	Relaxation	Intervention details N/R
Dimitrijevic et al., 2012			X	X	X	X			X			
Getz et al., 2012	X					X		X				
Jorgic et al., 2012	X		X	X		X						
Ballaz et.al., 2011			X	X		X					X	
Salem & Gropack, 2010				X	X	X			X	X	X	
Fragala-Pinkham et al., 2010			X	X		X				X		
Chrysagis et al., 2009			X		X					X		
Kelly et al., 2009				X	X	X			X	X		
Retarekar et al., 2009			X	X	X	X				X		
Fragala-Pinkham et al., 2009		X	X	X	X	X	X			X		
Fragala-Pinkham et al., 2008			X	X		X				X		
Aidar et al., 2007												X
Getz et al., 2007	X					X		X				
Őzer et al., 2007			X	X		X	X			X		
McManus et al., 2007				X		X	X					
Figuers, 2005		X		X					X			
Thorpe et al., 2005				X	X	X	X			X		

Note. When more than one intervention types were reported within an aquatic programme, X represents the main content of the programme.

2.2.4 Assessed outcomes

Twelve of the included research articles measured an outcome in the category of body functions and structures. For the category of activity and participation at least one outcome was measured in 15 research articles, whereas only three articles investigated an outcome in the category of personal factors. No outcomes of the environmental category were assessed (Table 2.3).

Table 2.3 The number of articles assessing at least one outcome in the components of the International Classification of Functioning (ICF), Disability and Health subdivisions, and quality of life.

ICF components	*Number of studies*
Body function and structure	12
Activity & Participation	15
Personal factors	3
Environmental factors	0
Quality of life	0

2.3 The effect of aquatics on the complex set of problems

The following sections present the effects of aquatics on the various issues that youth with CP encounter (pain, fatigue, walking ability, coordination, functional independence, QoL, self-esteem, and participation), the improvement of aquatic skills, and the enjoyment of the programmes, based on the systematically reviewed literature published since 2000.

2.3.1 Pain, fatigue and walking

Walking ability was investigated in various aquatic case studies (Fragala-Pinkham et al., 2009; Mackinnon, 1997; Retarekar et al., 2009; Salem & Gropack, 2010), in a few case series designs (Ballaz et al., 2011; Fragala-Pinkham et al., 2008; Thorpe et al., 2005) and in one controlled trial (Getz et al., 2012). Gait velocity was found to improve after a 4-month Halliwick programme in six 3 to 6 year-old children with CP (Getz et al., 2012) as both self-selected and maximum walking velocity over the 10-metre walk test improved significantly. Similarly, Fragala-Pinkham et al. (2008) reported a significant reduction in time for the half mile walk/run test after a 14-week aquatic exercise programme in a group of 16 children with disabilities. Additionally, self-selected gait velocity measured with the 3-minute walk test increased after a 10-week aquatic exercise programme in 6 of the 7 children with CP (Thorpe et al., 2005). However, in a sample of 10 adolescents with CP gait velocity did not improve after a 10-week aquatic programme (Ballaz et al., 2011). The metabolic cost of walking was found to decrease significantly in six 3 to 6 year-old children with CP after a 4-month Halliwick programme (Getz et al., 2012), and in 10 adolescents with CP after a 10-week aquatic programme (Ballaz et al., 2011). However, in a sample of 5 children with CP the metabolic cost of walking did not improve after 12 weeks of aquatic exercise (Kelly et al., 2009). The subdimension of the Gross Motor Function Measurement assessing walking, running and jumping abilities (scored with a Likert scale based on the quality of performance and the need for assistance) was reported to improve after all types of aquatic interventions (Ballaz et al., 2011; Chrysagis et al., 2009; Getz et al., 2012; Jorgić, Dimitrijević, Aleksandrović, et al., 2012; Thorpe et al., 2005). In summary, only one controlled trial was conducted to investigate the effect of aquatics on walking ability in children with CP that showed significant improvements in gait velocity and for the metabolic cost of walking in both the aquatic and the land-based intervention groups (Getz et al., 2012). The authors conducting case series designs reported no conclusive results for the effect of aquatics on these parameters.

Perceptions of fatigue did not increase or decrease after a 12-week aquatic group exercise programme in a group of five 9 to 11 year-old children with CP with the ability to walk (Kelly et al., 2009). In no other study the effect of aquatics on fatigue was assessed. The effect of an aquatic intervention on pain in youth with CP was not assessed in any of the identified studies. In summary, fatigue and pain are thought not to increase due to swimming; however, no literature can support this so far.

To conclude, no randomised controlled trial that was conducted to investigate the effect of swimming on walking velocity, pain and fatigue in children and adolescents with CP was retrieved in the literature.

2.3.2 Functional independence and coordination

In 5 of the 17 identified articles functional independence was assessed, all with the Pediatric Evaluation Disability Inventory (PEDI). Using a controlled trial, Getz et al. (2007) reported a significant improvement for functional independence in social functioning of 3 to 6 year-old children after a 4-month Halliwick programme. In a case series design including adolescents with CP who were not able to walk, a significant improvement for functional independence in social functioning was reported after a 16-week aquatic programme (Aidar et al., 2007). Although the results were not significant, an improvement for functional independence in mobility and self-care was noted after a controlled trial with a 4-month Halliwick programme for young children with CP (Getz et al., 2012). Similarly, in a case series study including children with mixed disabilities (Fragala-Pinkham et al., 2008) and a case study including a child with CP (Fragala-Pinkham et al., 2009), functional independence in mobility improved non-significantly after aquatic exercise programmes. Only in two of the five studies a comparison group was included, so the results need to be treated with caution. However, the literature suggests that an improvement in social functioning can be expected after an aquatic programme.

The effect of an aquatic programme on coordination using a general motor proficiency test was not studied yet. However, manual ability, measured with a paper and pencil task, was found to improve significantly in youth with CP without the ability to walk after a 16-week aquatic intervention (Aidar et al., 2007). Gross motor function (lower limbs and trunk), measured with the Gross Motor Function Measurement, was repeatedly found to improve (significantly and non-significantly) after various aquatic interventions ranging from 6 weeks to 4 months (Ballaz et al., 2011; Chrysagis et al., 2009; Dimitrijević et al., 2012; Getz et al., 2012; Jorgić, Dimitrijević, Aleksandrović, et al., 2012; Thorpe et al., 2005). These findings suggest that aquatic interventions can have a positive effect on coordination.

In conclusion, aquatic programmes aimed at youth with disabilities appear to have a positive effect on functional independence in mobility, self-care and social function. However, the effects of such programmes on functional independence have not been investigated using a RCT in a sample of children and adolescents with CP with the ability to walk. Coordination of the upper limb and bilateral coordination have not been evaluated after a swimming programme, despite the reported positive improvements in gross motor function and manual ability.

2.3.3 Self-concept, self-esteem and quality of life

Three of the 17 identified research articles addressed the effect of an aquatic intervention on self-esteem or self-concept. Body awareness improved significantly after a 14-week swimming intervention, compared to no swimming, in a randomised controlled trial including 23 children with CP aged 5 to 10 years (Őzer et al., 2007). Getz et al. (2007) conducted a controlled trial, consisting of a 4-month Halliwick programme in young children with CP, and reported non-significant improvements in perceived physical competence and social acceptance. However, social acceptance did improve significantly more than in the land-based intervention group (Getz et al., 2007). Thorpe et al. (2005) administered a 14-week aquatic resistive exercise programme and reported a trend towards improvement in self-perception in a group of children with CP, aged 7 to 13 years, but no comparison was made to a control group. The effect of an aquatic intervention on the QoL of youth with has not been investigated.

In summary, limited research supports the improvement in self-perception after swimming compared to no swimming. However, no adolescents were included in the samples. QoL in relation to an aquatic intervention was not evaluated.

2.3.4 Participation

Retarekar et al. (2009) and Fragala-Pinkham et al. (2010), using a case study and case series design respectively, reported that the parents perceived an increase in participation in physical activity after their children attended an aquatic exercise programme. Changes for participation in leisure activities as a result of an aquatic programme have not been evaluated in any other study.

2.3.5 Aquatic skills

Since 2000, the effect of an aquatic exercise, swimming or Halliwick-based intervention on aquatic skills of youth with neuromotor impairments was investigated in five studies. These include three case series designs (Fragala-Pinkham et al., 2008; Fragala-Pinkham et al., 2010; Jorgić, Dimitrijević, Aleksandrović, et al., 2012), one controlled trial (Getz et al., 2007) and one RCT (Dimitrijević et al., 2012). In the controlled trial and the RCT children aged 3 to 6 years (Getz et al., 2007) and 5 to 14 years (Dimitrijević et al., 2012) were included, and two different tools were implemented, namely the Aquatic Independence Measure (Getz et al., 2007) and the Water Orientation Test Alyn 2 (Dimitrijević et al., 2012). Significant improvements were evident after both the Halliwick (Getz et al., 2007) and the swimming (Dimitrijević et al., 2012) interventions. Unfortunately, none of the comparison groups performed the aquatic measurements and only Dimitrijević et al. (2012) included a follow-up period, which consisted of 3 weeks. These limitations make it difficult to draw a conclusion regarding the effect of a swimming intervention on swimming skills and the retention of these skills in youth with CP. In the studies using a case series design similar improvements over time were reported after an aquatic intervention (Fragala-Pinkham et al., 2008; Fragala-Pinkham et al., 2010; Jorgić, Dimitrijević, Aleksandrović, et al., 2012).

In summary, aquatic skills were evaluated in several intervention studies showing the effectiveness of an aquatic exercise or swimming programme on aquatic and swimming skills in children and adolescents with neuromotor impairments. However, neither a comparison with a control group was made, nor a long follow-up period was included, so no definite conclusion can be drawn on the effect of a swimming intervention.

None of the authors of all 17 studies reported the perceived level of enjoyment of the participants regarding the intervention programmes.

2.4 Conclusion

Since 2000, 17 research articles were published (in English), investigating the effects of an aquatic intervention in children and adolescents (6 months to 21 years old) with a disease or disability of the nervous system. Only six articles could be classified as having level of evidence I or II. No studies were published between 2000 and April 2013 that measured the effect of an aquatic intervention on pain, general motor proficiency and QoL in youth with neuromotor impairments. There is a lack of controlled studies (number of available controlled trials in parentheses) on fatigue ($n = 0$), walking ability ($n = 1$), functional independence ($n = 2$), self-perception ($n = 2$), aquatic skills ($n = 0$) and participation in leisure activities ($n = 0$) in children and adolescents with CP with the ability to walk. This is in line with the conclusion of the systematic review of Getz et al. (2006) including research articles until 2005. The number of published aquatic intervention studies in youth with neuromotor impairments increased in recent years, reflecting a growing interest in aquatics for youth with a disease of the nervous system. More importantly, the number of trials of high methodological quality increased.

A conclusion on the effectiveness of the various types of aquatic interventions is difficult to draw as programmes combined various types of interventions. Additionally, numerous outcomes were assessed, often using a variety of tools.

Nevertheless, aquatic programmes aimed at youth with neuromotor disabilities appear to have a positive effect on walking ability, functional independence, gross motor function and manual ability, self-perception, aquatic and swimming skills, and participation in physical activities. However, the methodological quality of the studies was low and various intervention types were used. Additionally, the two controlled trials (Getz et al., 2007; Getz et al., 2012) conducted to investigate walking ability (Getz et al., 2012), functional independence (Getz et al., 2007; Getz et al., 2012) and self-perception (Getz et al., 2007), included only 3 to 6 year-old children with CP. The RCT conducted to investigate self-perception included 5 to 10 year-olds, but only body awareness was assessed (Őzer et al., 2007). Kelly et al. (2009) reported no change over time for fatigue, using a case series design. The effect of aquatics on pain, coordination and QoL was not investigated in the identified literature. Therefore, it is necessary to undertake a RCT on the effects of swimming on pain, fatigue, walking velocity, upper limb and bilateral coordination, functional independence, self-perception, QoL and the participation in leisure activities, aquatic and swimming skills, in children and adolescents with CP. Additionally, the level of enjoyment of the programme needs to be assessed.

2.5 The current study; An RCT on the effects of aquatics in youth with CP

As described in Chapter 1, youth with CP encounter a complex set of issues interfering with physical, emotional and social well-being that contribute to the vicious cycle of deconditioning. A community-based swimming programme was proposed to have a positive effect on the issues, while the youth engage in physical activity. The literature review in Chapter 2 revealed a gap of controlled studies on the effects of swimming in children and adolescents with CP and the complex set of issues impacting physical, emotional and social well-being. Additionally, none of the authors reported the levels of enjoyment.

Therefore, the purpose of the present study was to investigate the effect of swimming on pain, fatigue, walking ability, functional independence, upper limb and bilateral coordination, self-perception, QoL, aquatic and swimming skills, as well as on participation in leisure activities, in youth with CP with the ability to walk. Additionally the retention of possible gains was investigated. Furthermore, the enjoyment of the swimming sessions was assessed, since motivation and enjoyment are essential to sustain a physically active lifestyle.

2.6 Research questions

The following section presents the research questions that were investigated. Figure 2.2 displays the relations between the issues in physical, emotional and social well-being that youth with CP encounter and the cycle of deconditioning, and displays the grouped variables addressed in Research Questions 1, 2, 3 and 6.

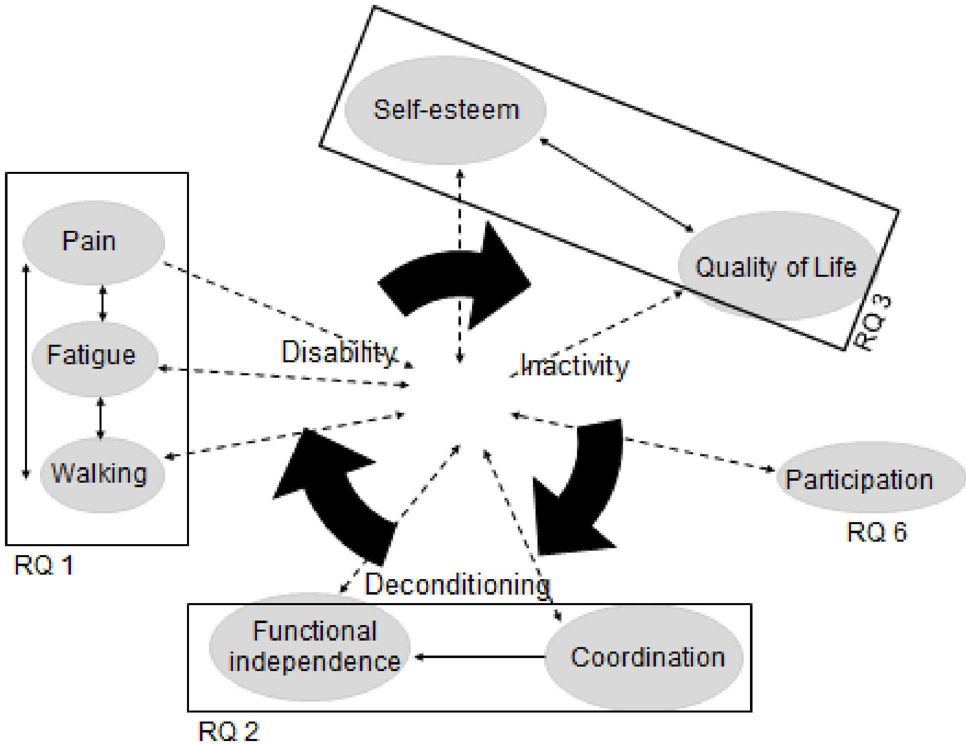


Figure 2.2 Multiple facets of functioning associated with the cycle of deconditioning, grouped per research question (RQ).

Research Question 1:
Deteriorated walking ability, pain and fatigue in youth with CP are associated with low physical activity and fitness levels. Swimming is a community-based exercise for which engagement and adherence to activity are expected to be facilitated. In the literature swimming has been stated to improve walking speed without increasing levels of pain or fatigue. However, due to the absence of a randomised controlled trial conducted to investigate the effect on walking speed, pain intensity and perceptions of fatigue in children and adolescents with CP, the following research question was investigated:

Does participating in a 10-week swimming programme affect walking ability, without adverse effects on pain intensity or perceptions of fatigue in youth with CP with the ability to walk?

Research Question 2:
Functional independence in daily living and general motor proficiency of the upper and lower limbs are key points of rehabilitation management in youth with CP. Low levels of physical activity and fitness partly can be explained by functional dependence and coordination difficulties. Swimming is proposed as a community-based exercise to combat the cycle of deconditioning as it encompasses learning the skill of swimming, which requires upper limb and bilateral coordination. Additionally, swimming in a community pool involves the training of functional independence in self-care, mobility and social functioning. However, due to the absence of a randomised controlled trial investigating the effect of swimming on functional independence and coordination in children and adolescents with CP, the following research questions was investigated:

Does participating in a 10-week swimming programme affect upper limb and bilateral coordination, and functional independence in youth with CP with the ability to walk?

Research Question 3:
Self-esteem, self-concept and quality of life have been reported being correlated and being lower in youth with CP than in their age-matched peers. Because of the positive effects of exercise on self-esteem, perceived competence and quality of life, swimming is proposed as an enjoyable exercise to affect perceived competence and quality of life, while youth are engaging in physical activity. Additionally, perceived sports competence is known to act as a mediator between motor proficiency and physical activity levels. Due to a lack of randomised controlled trials investigating the effect of swimming on perceived competence, global self-worth and self-reported quality of life, the following research question was investigated:

Does participating in a 10-week swimming programme affect perceived competence, global self-worth and self-reported quality of life in youth with CP with the ability to walk?

Research Question 4:
Perceived barriers reported by youth with CP to engaging in physical activity include the beliefs that learning a motor skill is too time-consuming and the perception of physical activity and sports as not being fun (Verschuren et al., 2012). Motor proficiency and enjoyment are important regarding the sustainability of a physically active lifestyle. Swimming is believed to be enjoyable and involves the learning of a skill that enables a person to engage in physical activities performed in the water. Swimming skills have been shown to improve after an aquatic intervention. However, no comparison with a control group has been made, nor a long follow-up period has been included, despite the importance of skill retention. Therefore, the following research question was investigated:

Does participating in a 10-week swimming programme affect mental adjustment in the water and swimming skills in youth with CP with the ability to walk; and can these improvements be retained over a 20-week follow-up period?

Research Question 5:

Fun and enjoyment are facilitators for engaging in physical activity, for improving adherence and for continuing committing to sport (Buffart et al., 2009; Martin, 2006; Redmond & Parrish, 2008). Therefore, physical activity and exercise programmes for youth with CP are recommended to be enjoyable and within the child's capabilities (Riner & Sellhorst, 2013). Swimming is believed to be fun; however, in none of the aquatic studies the levels of enjoyment of the interventions have been reported. Therefore, the following research question was investigated:

How is the 10-week swimming programme, offered to children and adolescents with CP, perceived in terms enjoyment?

Research Question 6:

Participating in community-based exercise and sport programmes could affect the low physical activity and fitness levels in youth with CP. However, participation in leisure activities has been reported being less varied and less intense in youth with CP than in their peers and with family close to home rather than with friends in the broader community. Learning to swim allows a person to engage in physical activities performed in the water. Swimming is an enjoyable community-based exercise that encourages sustaining a physically active lifestyle. Participation in physical activity has been reported to improve after an aquatic exercise programme in a case study and a case series design including children with mixed disabilities. Due to the absence of a trial including more than 2 participants with CP investigating the effect of swimming on participation in leisure activities, the following research question was investigated:

Does participating in a 10-week swimming intervention affect the diversity and intensity of participation in recreational, active-physical, social and skill-based activities, as well as with whom or where youth with CP participate?

Chapter 3: Methods

3.1 Participants

3.1.1 Inclusion and exclusion criteria

Flemish youth diagnosed with cerebral palsy (CP) aged 7 to 17 years were recruited to participate in the study. Inclusion criteria were the ability to walk indoors with or without hand-held mobility devices (Gross Motor Function Classification System levels I – III) and the ability to understand instructions. Exclusion criteria were a botulinum toxin A injection or orthopaedic surgery implemented or planned between 6 months prior to the start of the study and the end of the trial (Ward, 2008), or a medical contra-indication.

3.1.2 Recruitment

Participants were recruited through the 'CP reference centre' of a university hospital in the Flemish region of Belgium, via a specialist in rehabilitation medicine from another hospital in the Flemish region, via special schools, private practices and other contacts (such as previous participants, posters in swimming pools and in mainstream schools). The recruitment was conducted over a period of 6 months prior to the start of the study. The recruitment letter and poster are presented in Appendix B.

The board of the CP-reference centre was given an introductory presentation about the study. The doctors, physical therapists, psychologist and social worker of the team agreed on participation and a physical therapist was assigned responsibility for supervising the recruitment. All youth with a consultation appointment in the reference centre or the gait analysis lab in the 4-month period prior to the start of the study that met the inclusion criteria and lived within a radius of 100 km would be given information about the study on the planned consultation (+- 100 youth). Additionally, a recruitment poster was displayed in the hallway of the hospital's CP reference centre and the gait analysis lab. The specialist in rehabilitation medicine from another hospital agreed to ask the parents of the youth who met the inclusion criteria and showed interest in the study, to provide the main investigator with contact details. All were sent a letter and were contacted by phone afterwards.

A special school near the research centre agreed to forward information about the study to the families of youth who were eligible to participate. Twenty-five private physical therapists, in the area close to the research centre, were contacted by phone and 12 of these practices did not treat youth with CP. The other 13 private practices were provided with more information about the study, by e-mail or in person as they preferred. All therapists agreed to forward the information to youth that met the inclusion criteria. Every month the therapists were contacted by phone or e-mail for an update on whether youth and parents had shown an interest in the study. Subsequently, these families were provided with more information regarding the intervention study. Additionally, a poster was distributed to all personal contacts of the main investigator (e.g. other swimming instructors, participants of a previous intervention study, acquaintances) and of the co-investigator Professor D. Daly of the KU Leuven, Belgium. Posters were displayed at swimming pools and schools throughout the area of research centre.

3.1.3 Ethical approval

Before the start of the study, the parent(s) or guardian(s) provided full informed consent in addition to the verbal assent provided by the child or adolescent (Appendix C presents the informed consent form). Ethical approval was obtained from the Moray House School of Education Ethics Committee of the University of Edinburgh and from the University Hospital Medical Ethical Committee of the KU Leuven, where the study was conducted (Trial number: S53366).

3.2 Design

Research Questions 1 to 4 (presented in section 2.6) were investigated using a randomised controlled cross-over design. This design included two 10-week intervention periods, two 10-week control periods and two 5-week follow-up periods. The data of the perceived level of enjoyment regarding the swimming intervention (Research Question 5) were collected during the two 10-week intervention periods. Research Question 6, which aimed to assess the influence of swimming on the dimensions of participation, was investigated using a quasi-experimental one-group pre-test – post-test design. An outline of the study period is displayed in Figure 3.1.

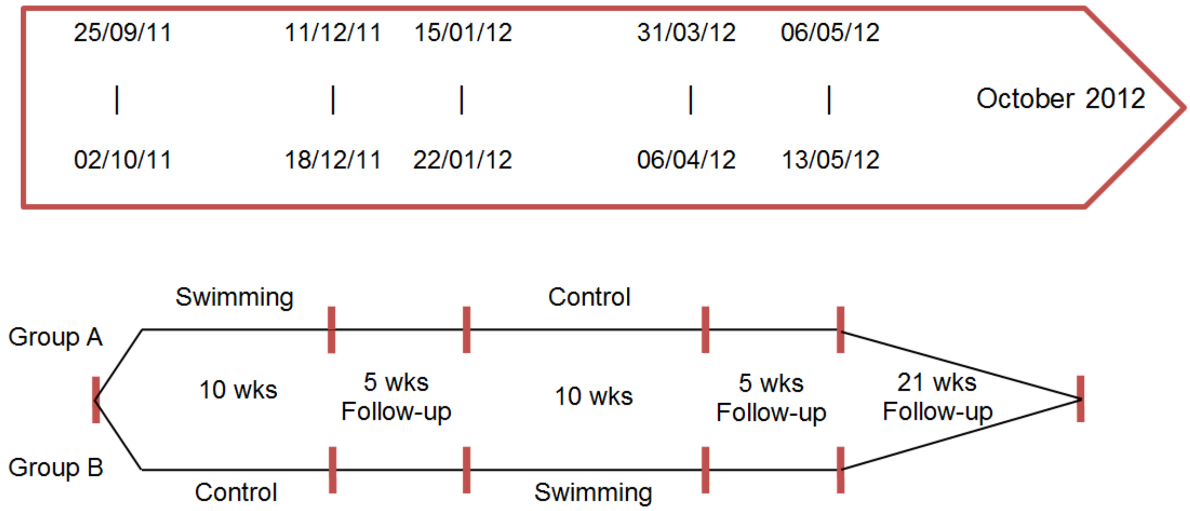


Figure 3.1 Timeline of the cross-over and the quasi-experimental design.

3.2.1 Randomised controlled cross-over design

Research Questions 1 to 4 were investigated using a randomised controlled cross-over design. An outline of the design is displayed in Figure 3.2. The participants were stratified and distributed randomly into two groups (process explained in section 3.3.1). After the baseline measurement, Group A participated in the swimming programme for 10 weeks, while Group B, acting as their control, received no extra therapy. A 5-week follow-up period with no scheduled swimming programme for either group followed. Group B, previously acting as the control group, participated immediately after this follow-up period in the 10-week swimming programme, while Group A received no extra therapy. Another 5-week follow-up period with no scheduled swimming programme for either group followed. All participants were evaluated five times: before and after both intervention/control periods and after both follow-up periods (T_1 to T_5, as displayed in Figure 3.2).

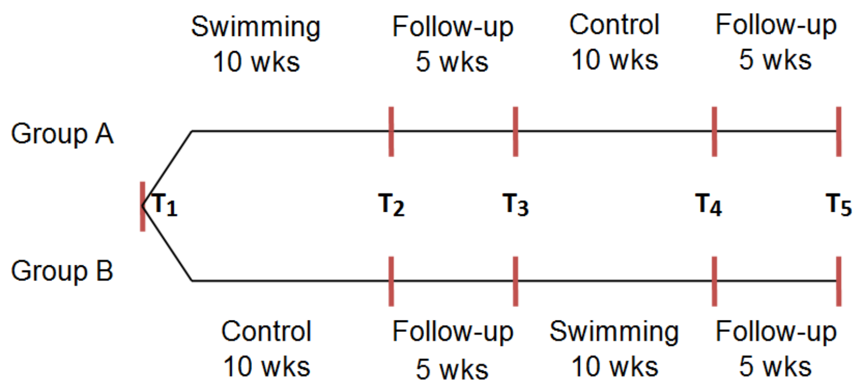


Figure 3.2 Study design for Research Questions 1 to 4.

A randomised controlled cross-over design is a strong scientific design and is marked as a high level of evidence and was therefore preferred. Additionally, a cross-over design was chosen to give all participants the opportunity to participate in the programme that was expected to benefit them. By maximising the number of youth entering the trial, this also maximises the amount of data gathered for the scientific analysis. However, data of the groups were not simply combined, as explained in section 3.6.2.

3.2.2 Quasi-experimental design

Research Question 6, which aimed to assess the effect of participating in the swimming intervention on the dimensions of participation, was investigated using a quasi-experimental one-group pre-test – post-test design (Figure 3.3). All participants were involved in the 10-week swimming programme for part of the 25 weeks following the baseline measurement. The timing of the swimming programme was not the same for all participants, as explained in the previous section (3.2.1). One year after the baseline measurement, all youth were re-evaluated with respect to their participation in leisure activities. Since all youth had participated in the 10-week swimming programme during the study period, there was no control group for this study. Therefore, a simple pre-test – post-test design was used.

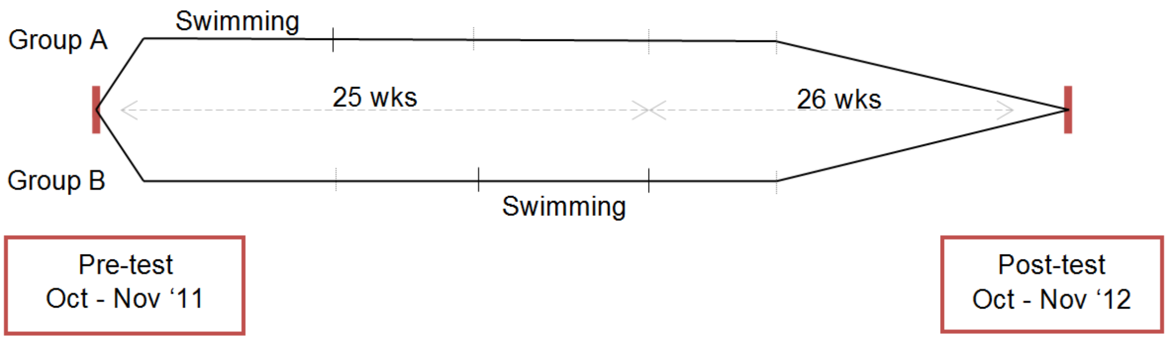


Figure 3.3 Study design for Research Question 6.

3.3 Procedures

3.3.1 Stratification and randomisation process

All participants were stratified before randomisation using two factors: three levels of motor function (Gross Motor Function Classification System (GMFCS) levels I, II, III and two levels of age (< 12.5 y; ≥ 12.5 y), resulting in a six-block randomisation. A block randomisation using sequentially numbered, opaque sealed envelopes (Doig & Simpson, 2005) and within-strata using two different block sizes (4 and 6) were used. Each step in the group allocation process was randomised (numbering the participants; allocation to two groups; sequence of intervention). The group allocation was performed by individuals who were not involved in the decision making at the inclusion process and who had no access to any information about the participants. A detailed description of the process is presented in Appendix D.

3.3.2 Evaluation procedure

All outcomes variables in relation to Research Questions 1 to 4 were measured on each measurement occasion (T_1, T_2, T_3, T_4 and T_5). This means that each participant performed the series of tests five times, with each testing session lasting approximately 1 hour and 30 minutes. All youth performed all measurements in the same order, except for the swimming test, which was performed before or after the series of outcome measurements (because it was performed at a different location). The order remained the same for each participant for all five measurement occasions. The order of the tests is presented in Appendix E. During testing, participants wore their own comfortable clothing, shoes and splints (as appropriate), and used their walking aid/s as appropriate. Parents completed a questionnaire by interview on the functional independence of their children (the scale is presented in section 3.5.2.2), which took 30 to 45 minutes; this was done by the same parent on each measurement occasion. Assessors were three Bachelors in Physiotherapy and Rehabilitation Sciences. At each measurement occasion, two of the three Bachelors assessed the participants and each assessed an equal number of participants of each group to avoid bias. The measurements in the water (Research Question 4) were assessed by the main investigator. Data on the level of enjoyment of the participants regarding the swimming sessions were collected after each swimming session. All these assessments were performed at the Faculty of Kinesiology and Rehabilitation Sciences of the KU Leuven and in the swimming pool where the intervention took place.

The questionnaire in relation to Research Question 6, aiming to assess participation in leisure activities, was distributed at the first measurement occasion (T_1). Youth were asked to complete the questionnaire at home during the week that followed. One year later, the same questionnaire was sent out by post, accompanied by an introduction and explanatory letter, and a return-envelope. The questionnaire can be completed in 30 to 45 minutes.

The assessors entered all data in Microsoft Excel twice, independently; hereafter Excel spread sheets were compared using Florencesoft^{TM} DiffEngineX^{TM}. Differences were highlighted and these numbers were compared with the original files. If scores had to be transformed, reversed or scaled, all procedures were performed twice, independently.

3.3.3 Blinding

Assessors tested the participants with no information concerning group allocation, and they were obliged to avoid questions or conversations on leisure activities or the intervention. Participants and their parents or guardians were aware of the blinding process and were asked to direct all questions to the main investigator. The main investigator who evaluated aquatic and swimming skills was aware of the group allocation. Participants were kept blinded to the goals of the study, but due to the nature of the intervention it was impossible to keep them blinded to the intervention or control condition. The swimming instructors were not kept blinded to the intervention or control condition either, but they were kept masked to the test results.

3.4 Intervention

The 10-week swimming programme in the community was offered without financial cost to the participants. The immediate objective of the programme was to teach the youth swimming skills, increasing their motor proficiency. For ethical reasons, all participants continued to receive their usual care and were not prohibited from swimming recreationally throughout the study period.

3.4.1 Quantity

The 10-week intervention consisted of two sessions per week of 30 to 60 minutes per session. This duration and frequency of intervention was chosen because significant improvements were reported for youth with CP on the body and activity component of the international classification of functioning, disability and health framework, after aquatic interventions with similar durations and frequencies in the systematic review by Jorgić, Dimitrijević, Lambeck, et al. (2012). Additionally, practical issues concerning family time and other leisure activities had to be kept in mind. For the children and adolescents attending the special school within walking distance from the swimming pool (Group A, $n = 4$ and Group B, $n = 3$), one session was performed during school hours. All other youth attended sessions outside school hours. When sessions were missed, extra sessions were organised to maintain the frequency of the planned intervention. The time spent in the pool and the number of completed sessions was noted. The median for each child was calculated and used for calculating the median attendance for each group. Individual compliance with the intervention had to be at least 60% for inclusion of the data in the analyses.

3.4.2 Content

The overall goal was to improve independence in the water (entry and exit of the pool, exhaling under water through mouth and nose, submerging the whole body, picking up objects under water, floating without assistance, transverse and longitudinal rotations, moving around freely in the shallow and deep end, jumping into the water, diving to the bottom, etc.) and to learn or improve a swimming stroke (breast stroke, front crawl and/or back crawl). All sessions consisted of a 5 to 10 minute warm-up with games and reviewing the tasks learned in the previous session, 20 to 40 minutes were dedicated to learning new tasks (the goals were set at the end of the previous session), and the session finished with 5 to 10 minutes of free play, races, and other games. Due to the variability in age, motor ability and swimming experience, all participants had an individual programme based on decision making models that are presented in Appendix F. A diary of all swimming sessions was kept for each swimmer to follow up progression and to adjust the individual goals for each session. Adolescents were involved in the goal setting of the programme, since adolescents reported to be keen to exercise their decision making skills, in contrast to being instructed what to do, which was reported as detrimental (Redmond & Parrish, 2008).

3.4.3 Intensity

The intervention programme was assessed in terms of exertion. Participants were asked to rate their perceived exertion after each swimming session using the OMNI scale (11-point Likert scale) (Lamb, Parfitt, & Eston, 2008, p. 149; Robertson et al., 2000). The scale is presented in Appendix E. The OMNI scale has been found valid for use in typically developing children performing walking or running exercises (Robertson et al., 2000; Utter, Robertson, Nieman, & Kang, 2002) and it has been found valid and reliable for use in typically developing adolescent girls in submaximal exercise conditions (Pfeiffer, Pivarnik, Womack, Reeves, & Malina, 2002). No studies have been performed to test validity and reliability of the OMNI scale for use in swimming. For each participant, the median and the inter quartile range (IQR) of the scores that were reported for each swimming session were calculated. These medians were used to calculate the medians and IQRs for Group A and for Group B.

3.4.4 Location

The intervention was conducted in a 25 m by 13 m swimming pool at the KU Leuven, Belgium, which had a temperature of 27 – 28°C. The swimming sessions took place in both the shallow end, stretching the first 13 m of the pool's length with a depth of 125 cm to 135 cm, and in the deep end.

3.4.5 Instructors

Participants were individually tutored and some activities were carried out with the entire group present in the pool (maximum of 4 participants). The main investigator instructed the youth in the pool assisted by students from the Faculty of Kinesiology and Rehabilitation Sciences of the KU Leuven.

3.5 Measures

3.5.1 Descriptive variables

The following information was gathered prior to the baseline measurement: date of birth, gestational age, GMFCS level, Manual Ability Classification System level, amount of physical therapy on the current rehabilitation programme, use of orthotic devices, swimming experience and type of education. Parents and the physical therapists provided information concerning the type of CP and topography, and related medical problems. The participants' height and weight were measured at the five measurement occasion of the cross-over design (Figure 3.2). During the study period the parents and youth were asked to keep a diary of the gross motor leisure activities (e.g. cycling), upper limb leisure activities (e.g. playing music) and changes in the amount of physical therapy per week.

3.5.2 Outcome variables

3.5.2.1 Visual Analogue Scale, Faces Pain Scale - Revised, 10-metre walk test, 1-minute fast walk test, PedsQL™ multidimensional fatigue scale

Self-reported pain intensity was measured using the Faces Pain Scale - Revised (FPS-R) (Hicks, von Baeyer, Spafford, van Korlaar, & Goodenough, 2001) and the Visual Analogue Scale (VAS). Both tools are valid and reliable and are considered the most appropriate tools for use in clinical trials in children (FPS-R) and adolescents (VAS) (Stinson, Kavanagh, Yamada, Gill, & Stevens, 2006; Tomlinson, von Baeyer, Stinson, & Sung, 2010). The version of the VAS that was used in the present study was a ruler with a 100 mm black horizontal line labelled with endpoints 'no pain' and 'worst imaginable pain'. Each participant was asked to slide the indicator to a place on the line corresponding to their amount of hurt or pain (intensity) in the past week. The other side of the ruler showed the same line, with marks every millimetre, and the place of the indicator. The VAS was scored from 0 to 100, with 1 unit equivalent to 1 mm. As practice, participants were asked about a previous pain experience and the worst pain imaginable, and how to highlight these on the 100 mm line. The FPS-R shows six facial expressions, indicating 'no pain' to 'very much pain', which was explained to the youth. Each participant was asked to mark the face that reflected his or her current pain. The FPS-R is scored from '0' = 'no pain' till '10' = 'very much pain'. Words like 'happy' and 'sad' were not used. Both the VAS and the FPS-R were used for all participants, since the use of the FPS-R has been suggested for school-aged children (4 to 12 years old) and the use of the VAS for youth aged 8 years or older, especially in clinical trials (Stinson et al., 2006). However, for the latter the supplementary use of the FPS-R was recommended, particularly for children aged 8 to 12 years (Stinson et al., 2006).

Walking ability was measured using the 10-metre walk test (10-MWT) and the 1-minute fast walk test (1-min WT). The 10-MWT measures self-selected walking speed, and it is indicative of the distances youth commonly need to walk between classrooms, to the toilet, or within their home (Willoughby, Dodd, Shields, & Foley, 2010). The participants were instructed to walk at a comfortable pace over a

14-metre distance. Two meters at the start and at the end allowed for acceleration and deceleration. A stopwatch recorded the time it took to walk the middle 10 m. Shorter times are considered to reflect a better walking capacity. The mean speed of three attempts was calculated. This test has been found to be reliable and valid for use in adults with neurological disorders (Rossier & Wade, 2001; Van Hedel, Wirz, & Dietz, 2005). After the 10-MWT, participants were allowed a maximum of 5 minutes seated rest before starting the 1-min WT. The user-friendly 1-min WT was performed at the participant's maximum walking speed. At the start of the test, participants stood at a marked point inside the outline of a 20 m level oval track. They were told that when the instruction to start was given, they had to keep walking around the track as fast as possible for one minute, without running. The distance was calculated to the nearest metre using metre markings on the track. During the test, youth were told the elapsed time after 30 s and again when 10 s remained. The 1-min WT test is valid and reliable for children with CP (McDowell, Humphreys, Kerr, & Stevenson, 2009; McDowell, Kerr, Parkes, & Cosgrove, 2005).

Perceptions of fatigue was measured using the Dutch translation of the 'PedsQL™ multidimensional fatigue scale – standard version – Belgian Dutch' (PedsQL Fatigue) (Varni, Burwinkle, Katz, Meeske, & Dickinson, 2002; Varni, Burwinkle, & Szer, 2004; Varni & Limbers, 2008), which is available as a self-report scale for the ages 8 – 12 years (child), and 13 – 18 years (adolescent). This scale consists of 18 items and each item rates how often a particular problem occurred in the past month, using a 5-point Likert scale. Each item was reverse-scored and rescaled to a 0 – 100 scale, so that higher scores indicated fewer symptoms of fatigue. If 50% or more of the items were completed, a total score was calculated by adding up the scores for all items divided by the number of answered items. The Dutch translation is valid and reliable for use in children and adolescents (Gordijn, Cremers, Kaspers, & Gemke, 2011). Children completed the questionnaire together with the assessors; adolescents completed the questionnaire by themselves and could ask questions if required.

3.5.2.2 Bruininks-Oseretsky test of motor proficiency, Pediatric Evaluation of Disability Inventory

Bilateral and upper limb coordination were assessed with the subtests 4 and 7 of the Bruininks-Oseretsky test of motor proficiency (2nd edition) (BOT-2) (Bruininks & Bruininks, 2005). This scale is an individually administered assessment designed to measure fine and gross motor skills of children and adolescents aged 4 to 21 years. Subtest 4, *bilateral coordination*, contains seven items including touching the nose with the index fingers, jumping jacks, pivoting thumbs and index fingers, tapping feet and fingers. Subtest 7, *upper limb coordination*, contains seven items including catching, dribbling and throwing a ball. Raw item scores were recorded for each item. Two trials were allowed when appropriate (the record form is presented in Appendix E). Raw item scores were converted to point scores using information provided on the record form. For each subtest, the point scores were summed, creating total point scores (Bruininks & Bruininks, 2005) ranging from 0 to 24 and 0 to 39 for subtests 4 and 7 respectively. Subtest total point scores were converted to gender- and age- specific scale scores, with the age based on the examinee's chronological age in years and months (disregarding days) at the time of measurement (Tables B.1 – B.2 in Bruininks and Bruininks (2005)). Inter-rater reliability is known to be high for both subtests in children and adolescents with a variety of disabilities aged 4 to 21 years (Bruininks & Bruininks, 2005). The test-retest reliability for both subtests in children and adolescents varies from 0.48 to 0.71 (Bruininks & Bruininks, 2005). Both subtests show test-retest reliability scores > 0.85 when tested in children with intellectual disabilities (Wuang & Su, 2009). Both subtests have been found to be valid for use in children and adolescents with a variety of disabilities aged 4 till 21 years (Bruininks & Bruininks, 2005; Deitz, Kartin, & Kopp, 2007).

Functional independence was assessed using the Dutch version of the Functional Skills Scale of the 'Pediatric Evaluation of Disability Inventory' (PEDI-NL) (Custers, Wassenberg-Severijnen, et al., 2002; Haley, Coster, Ludlow, Haltiwanger, & Andrellos, 2005). This scale is a parent-report by interview and contains 205 items across three subscales measuring daily living skills in the functional domains of self-care, mobility and social function. Each item was scored dichotomously: either 'capable' or 'unable' to perform the task in most situations. Raw subscale scores were calculated for each subscale by adding up the items, and were transformed to a 0 – 100 scaled score, with a higher score thus indicating greater capability (Haley et al., 2005). Scaled scores give an indication of functional performance regardless of age. The PEDI is designed for detecting functional deficits, to monitor progress and to act as an outcome measure in children aged 6 months to 7.5 years. It may be used to assess children at older chronological ages who demonstrate deficits in basic functional activities (Custers, Wassenberg-Severijnen, et al., 2002). Youth with CP show these deficits and the scale has been used in youth with CP aged up to 18 years, covering all GMFCS levels (Kerr et al., 2011; Ohata et al., 2008; Öhrvall et al., 2010). However, the chance of a ceiling effect exists when using the test with older children and adolescents. The PEDI is sensitive to small changes (Ziviani et al., 2002) and psychometric properties of the PEDI-NL have been reported to be good (Custers, van der Net, et al., 2002; Custers, Wassenberg-Severijnen, et al., 2002; Wassenberg-Severijnen, Custers, Hox, Vermeer, & Helders, 2003).

3.5.2.3 Self-Perception Profile for youth with CP, PedsQL™ cerebral palsy module version 3.0

Self-worth and perceived competence were assessed using the '*Competentie Belevingsschaal voor Kinderen/Jongeren met Cerebraal Parese*' (CBSK-CP) (Komdeur, Schuur, Wijnroks, & Vermeer, 2001; Veerman, Straathof, Treffers, Van den Bergh, & ten Brink, 1997). This scale has been derived from the Dutch version of Harter's self-perception profile for children/adolescents scale, as adapted by Komdeur et al. (2001) for use with children and adolescents with CP aged 9 to 16 years. The CBSK-CP is a 49-item scale consisting of five domain-specific subscales (scholastic competence, social acceptance, athletic competence, physical appearance and motor competence) and one subscale that measures global self-worth. The scale makes use of a structured alternative format. For each item, participants were asked to choose from two opposite statements that statement they believed described them the best and to indicate whether the statement was somewhat true or very true. For example: 'Some kids often forget what they learn – Other kids can remember things easily': the participant was asked to choose the statement they believed applied best

to them and to indicate whether that statement was 'really true' or 'sort of true' for them. The first item was for practice and was not included in the calculations. The items on each question were scored from one to four, where one is construed as a low perceived competence and four indicates a high perceived competence.
Domain-specific scores were calculated by adding up the scores of the eight items within each domain. Each of these subscales' scores ranged from 8 to 32. The mean of the subscores for each subscale was used for further analyses. The global self-worth subscale consists of eight items, independent from the other subscales. An overall score for the CBSK-CP is not provided. This tool has been found valid and reliable for use in children and adolescents with CP (Komdeur et al., 2001).

Self-reported physical functioning and related quality of life were evaluated using the Dutch translation[a] of the validated 'PedsQL™ cerebral palsy module version 3.0' (PedsQL CP) (Varni et al., 2006), which is available as a self-report scale for the ages 8 – 12 years (child), and 13 – 18 years (adolescent). This scale is composed of 35 items comprising seven dimensions (daily activities, school activities, movement and balance, pain and hurt, fatigue, eating activities, and speech and communication). Each item rates how often a particular problem occurred in the past month using a 5-point Likert scale. Each item was reverse-scored and rescaled to a 0 – 100 scale, so that higher scores indicated a higher quality of life. Scores were computed for each dimension as the sum of the item scores divided by the number of items answered. If more than 50% of the items in the dimension were missing, the dimension score was not computed. No total score was computed (Varni et al., 2006). Internal consistency has been demonstrated for the CP module and construct validity has been established for the English version of the questionnaire (Varni et

[a] A Belgian-Dutch translation was developed prior to the start of the study according to the Linguistic Validation Guidelines provided by the Mapi Research Institute, including a forward and backward translation and a patient testing phase. Two native target language speakers (including the main researcher) made an individual and independent forward translation of both the parent-report and self-report forms for the age groups 8 to 12 and 13 to 18 years old, into the target language, Dutch. One native US English speaker used the first Dutch version to make a backward translation into the original language, English, without access to the original US English version of the questionnaire. The backward translation was compared with the original version of the questionnaire. The patient testing phase included 5 children and 5 adolescents and their parents, for the respective questionnaire type.

al., 2006). Children completed the questionnaire together with the assessors; adolescents completed the questionnaire by themselves and could ask questions if required.

3.5.2.4 **Water Orientation Test Alyn 2**

Aquatic and swimming skills were evaluated using the Water Orientation Test Alyn 2 (WOTA 2), which is a 27-item test based on the Halliwick concept (International Halliwick Association Education and Research Committee, 2010) that assess the swimmer's level of adjustment and function in the water (Tirosh, Katz-Leurer, & Getz, 2008). It consists of two parts: a 13-item 'mental adjustment' part (MA) and a 14-item 'skills, balance control and movement' part (SBM). Both the swimmer and the instructor were in the water at the time of testing. In addition to giving verbal instruction, the instructor demonstrated the task to be performed and each item was attempted up to three times. A 4-point ordinal scale (0 – 3) was used to score each skill based on the level of performance and independence. A maximum of 81 points could be obtained and all scores were converted to a percentage of the maximum. The scale has been found to be reliable and valid for use in children and adolescents with disabilities (Tirosh et al., 2008).

3.5.2.5 **Enjoyment of the intervention**

The rating of the level of enjoyment regarding the swimming intervention (5-point Likert scale) was taken after each swimming session. The scale is presented in Appendix E. No studies were performed to test the validity and reliability of the 5-point Likert scale rating the level of enjoyment, however, the same scale is used in the validated Children's Assessment of Participation and Enjoyment.

3.5.2.6 Children's Assessment for Participation and Enjoyment

Five participation dimensions were assessed using the Dutch version of the Children's Assessment of Participation and Enjoyment (CAPE) (King et al., 2010). The CAPE consists of 55 activities and measures participation in formal (structured, involving rules or goals) and informal (spontaneous, involving little planning, initiated by the child) leisure and recreation activities outside school for youth aged 6 to 21 years. This scale is a self-report and was completed by the participant assisted by their parent/guardian when necessary. All 55 activities are illustrated and five dimensions of participation are measured: (1) diversity: 'Have you done this activity in the past 4 months?' (yes/no); (2) intensity: 'How often?' (7-point Likert scale, ranging from once in the past 4 months, to at least once per day); (3) with whom: 'With whom do you do this most often?' (5-point Likert scale, ranging from alone, with family, friends to with others); (4) where: 'Where do you do this most often?' (6-point Likert scale, ranging from at home, in the neighbourhood, in town, to out of town) and (5) enjoyment: 'How much do you like or enjoy doing this activity?' (5-point Likert scale using smiley faces and labels ranging from 'not at all', 'a little', to 'very much'). A score for each dimension was obtained for all activities together (total scores), for the informal and formal domain, and for the five activity types. The formal domain consists of 15 items and the informal domain of 40 items. The 55 items across five activity types are recreational (12 items), active-physical (13 items), social (10 items), skill-based (10 items) and self-improvement (10 items) activities. The Dutch version of the scale has been found valid and reliable to measure participation in recreation and leisure activities for youth with physical disabilities aged 6 to 18 years (Bult et al., 2010).

3.6 Data analysis

3.6.1 Sample size calculation

Sample sizes were calculated based on an α-level of .05, a statistical power of .80, expected effect sizes (ES) of 0.80 for the WOTA 2 and 0.50 for all other outcome measures, and the statistical test expected to use. The ES were chosen based on the conventional ES reported by Cohen (1988) suggesting *d* values of 0.8, 0.5 and 0.2 represent large, medium and small ES. G*Power 3, a statistical power analysis program (Faul, Erdfelder, Lang, & Buchner, 2007) was used.

For the present study, the analysis of WOTA 2 required a sample size of 26 in each group when using an independent *t*-test, to achieve statistical power of .80. The analyses for all other outcome measures related to Research Questions 1 to 4 required sample sizes of 64 in each group when using an independent *t*-test. The CAPE required a sample size of 34 participants when using a dependent *t*-test, to achieve statistical power of .80.

3.6.2 Statistical analysis of the randomised controlled cross-over design

Research Questions 1 to 4 were investigated using a randomised controlled cross-over design (Figure 3.2). Normally, these designs are analysed by pooling the data of both groups (Hopkins, 2000). As the present study involved a skill-training intervention, a carry-over effect was expected, and therefore the data were not pooled (Hopkins, 2000). It is possible to estimate the carry-over effect as the first step in a two-step approach to the analysis of this type of data (Shen, 2006), but principally other literature criticises this two-stage analysis as being misleading, and therefore advises against its use (Freeman, 1989; Ratkowsky, Evans, & Alldredge, 1993; Senn, 1993, 2006). In spite of the expected carry-over effect, a cross-over design was used for ethical reasons, that is, participants in Group B, firstly serving as a control group, were given the opportunity to participate in the swimming intervention to gain the potential benefits (waitlist control). Also, given the small number of participants, the additional data of Group B were used to assess agreement with the main analysis' outcome (the RCT indicated in Figure 3.4a), using a within-group comparison

(Figure 3.4b). A further advantage of the design was that the 20 weeks in which Group A did not receive the swimming intervention, enabled assessment of whether the changes in the outcome variables were retained after a 15- and 20-week follow-up period (Figure 3.4c).

As pooling of the data was not appropriate, the main analysis used the data of the first fifteen weeks of the study that included measurement occasions T_1, T_2 and T_3. This analysis included an intervention and control group receiving a swimming and no additional therapy programme, respectively, and included a 5-week follow-up period. This part of the study thus constitutes the Randomised Controlled Trial (RCT) - part (Figure 3.4a). Figure 3.4b shows which data sets were used to assess agreement with the main analysis, using a within-group comparison (BA-design) that includes a control and swimming period for Group B. Finally, Figure 3.4c shows which data sets were used to assess the retention of the changes in Group A over a 20-week period, including measurement occasions T_1, T_4 and T_5.

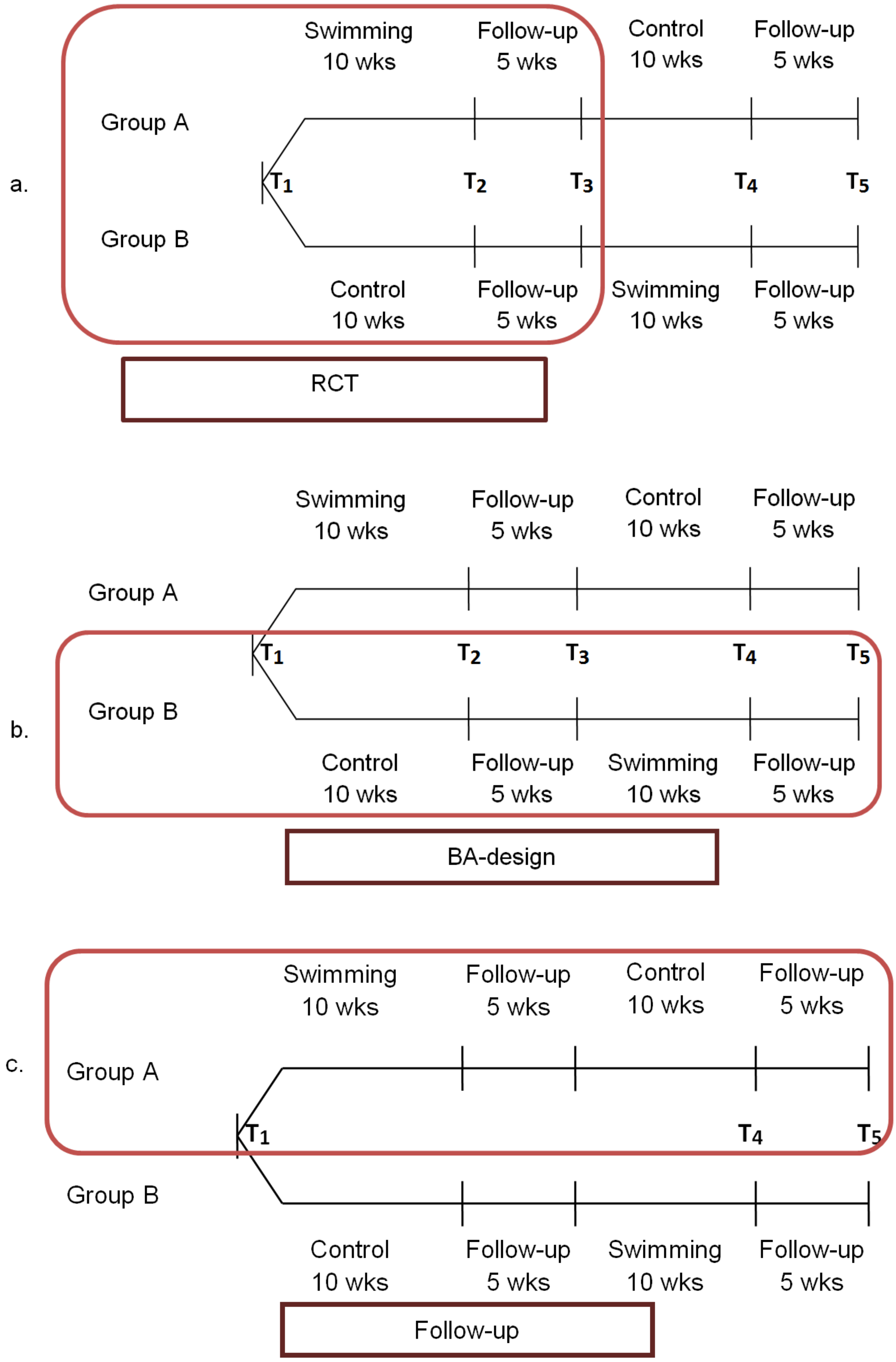


Figure 3.4 Statistical analysis of the cross-over design; a. Main statistical analysis: Randomised Controlled Trial (RCT); b. Statistical analysis to assess agreement with results of main analysis; c. Statistical analysis to assess the retention of the changes.

Normality of the baseline values of all tests for each group was tested using Shapiro-Wilk tests in SPSS v19 (Table 3.1). If data appeared to be distributed non-normally, square root transformations were applied and normality was tested subsequently. Data analyses were performed using SPSS v19 and Microsoft Excel 2010, and the α-level was set at .05. The exact (two-tailed) *p*-value was used in the non-parametric analysis, as the exact calculation produces a reliable result regardless of the size, distribution, sparseness or balance of the data (Mehta & Patel, 1989, 2010).

The main analysis (Figure 3.4a) included T_1, T_2 and T_3, constituting a RCT comparing swimming with no swimming for two independent groups. Comparability of the groups at baseline was assessed using Mann-Whitney U tests for the ordinal and continuous descriptive measures and a chi-square test for the categorical descriptive measures (gender, swimming experience (y/n), education type, use of orthotic devices (y/n)). As the expected cell count was lower than five, the Fisher Exact test values were used (Munro, 2005). The baseline values of the outcome variables were compared between Group A and B using a Mann-Whitney U test or independent *t*-test as appropriate.

Non-parametric analyses were used to analyse pain intensity as the FPS-R is a 6-point Likert scale and the VAS is a 0 – 100 mm scale, representing the continuum of pain, and for each measurement the data were not normally distributed in at least one group at baseline (Table 3.1). The PedsQL Fatigue is an 18-item 4-point Likert scale and the baseline values of Group B were not normally distributed (Table 3.1), therefore non-parametric analyses were used to analyse the perceptions of fatigue. The two walking tests (10-MWT, 1-min WT) involve ratio scales that measure velocity and distance respectively. For both scales the baseline values of each group were normally distributed (Table 3.1) and the homogeneity of variance was confirmed with the Levene's test, so parametric analyses for both walking tests were used.

Non-parametric analyses were used to analyse the BOT-2 and the PEDI-NL as the bilateral coordination subitem and the social function subscale were not normally distributed at baseline (Table 3.1).

Parametric analyses were used to analyse the CBSK-CP as the subscores at baseline had a normal distribution (Table 3.1) and the homogeneity of variance was confirmed with the Levene's test. As the PedsQL CP includes 4- to 9-item 5-point Likert scales and was not normally distributed at baseline in four dimensions (Table 3.1), non-parametric analyses were used.

Non-parametric analyses were used to analyse the WOTA 2 as the baseline values of Group A had a non-normal distribution (Table 3.1) and this scale involves 27 items on a 4-point Likert scale. All analyses were performed for the total score, the MA subscore and the SBM subscore, to see if changes occurred for the swimming skills in general and for the subscales.

Since none of the data that was distributed non-normally appeared to be distributed normally after the square root transformation, raw data was used in further data analyses.

Table 3.1 Results of the Shapiro-Wilk tests for all outcome variables at baseline for both groups.

Outcome variable		*p* Group A	*p* Group B
Water Orientation Test Alyn 2	Total score	.018*	.507
	MA subscore	.006**	.296
	SBM subscore	.696	.972
Visual Analogue Scale		.031*	.089
Faces Pain Scale–Revised		.004**	.005**
10 metre walk test		.082	.172
1-minute fast walk test		.268	.567
PedsQL™ multidimensional fatigue scale		.577	.009**
Bruininks-Oseretsky test of motor proficiency 2nd ed.	Bilateral coordination	.162	.016*
	Upper limb coordination	.549	.963
Pediatric Evaluation of Disability Inventory	Self-care	.325	.843
	Mobility	.180	.381
	Social Function	.174	.014*
Self-Perception Profile for Youth with CP	Scholastic competence	.133	.624
	Social acceptance	.379	.498
	Athletic competence	.596	.780
	Physical appearance	.699	.676
	Motor competence	.738	.667
	Global self-worth	.378	.872
PedsQL™ CP module (3.0)	Daily activities	.927	.646
	Social activities	.045*	< .001**
	Movement & balance	.021*	.055
	Pain & hurt	.314	.236
	Fatigue	.159	.264
	Eating activities	.314	.006**
	Speech & communication	.042*	.007**
Children's Assessment for Participation and Enjoyment	Diversity	.368	
	Intensity	.253	
	'With whom'	.657	
	'Where'	.166	
	Enjoyment	.240	

Note. MA: mental adjustment, SBM: skills, balance control and movement. * $p < .05$; ** $p < .01$.

Differences between groups over time (RCT design; Figure 3.4a) were analysed using Mann-Whitney U tests comparing the groups' change scores over the 10-week period (T_1–T_2)[b], and comparing the groups' change scores over the 15-week period (T_1–T_3)[c], for the pain intensity scales, the PedsQL Fatigue, both subscales of the BOT-2, all three subscales of the PEDI-NL, all subscales of the PedsQL CP and the WOTA 2. For all these outcome measurements, the changes over time (T_1, T_2, T_3) were analysed using a Friedman's Two-way Analysis of Variance by ranks for each group separately. Where significant results were found with the Friedman's Two-way ANOVA by ranks, post hoc tests suggested by Siegel and Castellan (1988) (Bull et al., 2004; Pett, 1997) were used to test the differences between baseline (T_1) and post-test (T_2) and between baseline (T_1) and follow-up test (T_3). These tests corrected for multiple comparison. For both walking tests and the CBSK-CP subscales, differences between groups over time were analysed using independent *t*-tests comparing the groups' change scores over the 10-week period, and comparing the groups' change scores over the 15-week period (Hopkins, 2003). Confidence intervals (95%) are reported. The changes over time were analysed using a repeated measures ANOVA for each group separately (simple contrasts, first). When assumptions of sphericity were not met, the Greenhouse Geisser correction was used (indicated in the results with GG). Bonferroni correction for multiple testing was applied.

To assess agreement with the results, an additional within-group analysis was performed for Group B, acting as their own control (BA-design) (Figure 3.4b). T_5 was excluded from the analysis of Group B for all outcome variables as 4 of the 7 participants of Group B had a botulinum toxin A injection and/or a limb in a cast between T_4 and T_5 (as described in results section 4.4). A Wilcoxon signed-rank test was used to compare change scores over the intervention period (T_3–T_4)[d] with change scores over the control period (T_1–T_2) for both pain intensity scales, the PedsQL Fatigue, both subscales of the BOT-2, all three subscales of the PEDI-NL, all

[b] T_1–T_2 represents the period between T_1 and T_2 and is calculated as T_2 minus T_1.
[c] T_1–T_3 represents the period between T_1 and T_3 and is calculated as T_3 minus T_1.
[d] T_3–T_4 represents the period between T_3 and T_4 and is calculated as T_4 minus T_3.

subscales of the PedsQL CP and the WOTA 2. For both walking tests and the subscales of the CBSK-CP, paired-samples *t*-tests were used to compare changes over the intervention period with changes over the control period.

To assess whether these gains were retained over the 15- and 20-week follow-up period that followed the completion of the intervention programme in Group A (Figure 3.4c), Wilcoxon signed-rank tests and paired-samples *t*-tests were used to test Group A for differences between T_1 and T_4 and between T_1 and T_5. The outcome variables analysed with non-parametric statistics, as described above, were analysed with Wilcoxon signed-rank tests and the outcome variables analysed with parametric statistics were analysed with paired-samples *t*-tests.

Non-parametric data are visually presented in the results, using boxplots that represent the median, the upper and lower quartiles, and the whiskers. Outlying scores are plotted as individual points.

3.6.3 Analysis of the perceived enjoyment of the intervention (Research Question 5)

The intervention programme was assessed in terms of enjoyment. Participants were asked to rate their perceived level of enjoyment after each swimming session. For each participant the median and the IQR of the scores reported for each swimming session were calculated. These medians were used to calculate the medians and IQRs for Group A and Group B separately.

3.6.4 Statistical analysis of the quasi-experimental design

Research Question 6 was investigated using a one-group pre-test – post-test design. Changes in participation intensity, diversity and enjoyment, and with whom and where activities were performed, were analysed for the total score, the informal and formal domain, and the five activity types. The diversity score represents the amount of activities that was participated in during the past 4 months. The percentage of diversity was this amount of activities divided by the maximum possible score of 55, multiplied by 100. Intensity represents the mean of the frequency of participation for all 55 activities during the past 4 months. All other scores (with whom, where and enjoyment) represent the mean scores for the activities that had been participated in during the past 4 months. Since baseline data were normally distributed (Table 3.1), paired-samples *t*-tests were used to compare the results at baseline and the results one year after baseline.

3.6.5 Effect sizes

Effect sizes (ES) for Mann–Whitney U and Wilcoxon signed-rank tests were calculated as follows: $r = Z / \sqrt{N}$ (Fritz, Morris, & Richler, 2012). Cohen's guidelines for *r* suggest that values of 0.5, 0.3 and 0.1 represent large, medium and small ES respectively (Cohen, 1988; Coolican, 2009, p. 395).

ES for the parametric tests were calculated as the difference between the means divided by the pooled sample standard deviation, corrected for bias (Fritz et al., 2012). Confidence intervals (95%) for these ES are reported. These ES were interpreted using the guidelines from Cohen (1988) suggesting that *d* values of 0.8, 0.5 and 0.2 represent large, medium and small ES respectively.

3.6.6 Clinical analysis

As statistical significance is dependent on sample size, a minor change in a large group of participants can be statistically significant and a major change in a very small group may not reach significance. In other words, significance does not automatically imply a clinically relevant result, and a lack of significance does not automatically imply that there is no clinical relevance to the results. Hence, clinical relevance needs to be judged in addition to significance (Portney & Watkins, 2000, p. 410).

For the 1-min WT the repeatability coefficient was reported being 13 m in the test-retest reliability study of McDowell et al. (2009) in youth with CP with GMFCS levels I to III (aged 3 to 18 years). Therefore, changes larger than 13 m for the 1-min WT were deemed clinically relevant. Lam, Noonan, Eng, and Team (2008) calculated that the smallest detectable change with 95% confidence for the 10-MWT is 0.13 m/s in adults with spinal cord injury.

Changes larger than 10 mm on the VAS are reported to represent a significant change in acute pain (Stinson et al., 2006). The clinically important difference for chronic pain in youth, measured using the VAS, has not been reported. Von Baeyer (2006) reported that a change of one face for the FPS-R (representing a change of 2 points) is the estimate of the minimum clinically significant difference in children.

Wuang and Su (2009) reported that the minimal important differences between ‘no change’ and ‘improved’ for the BOT-2 are 1.1 and 1.6 for the bilateral coordination subtest and the upper limb coordination subtest, respectively, in a sample of youth with an intellectual disability.

The manual of the PEDI reports that differences larger than twice the standard error reported in the manual, are clinically significant (Haley et al., 2005). Standard errors for the scaled scores of the PEDI-NL have been reported to be between 0.7 and 2.1.

According to Shields et al. (2007), a mean score greater than 2.5 indicates a positive self-concept, whereas a mean score lower than 2.5 indicates a negative self-concept for the subscales of the self-perception profile for children/adolescents. The rationale for this cut-off is based on the fact that 2.5 is the midpoint between the highest negative self-judgement (score of 2) and the lowest positive self-judgement (score of 3).

The minimal detectable change based on the confidence interval of 95% for the WOTA 2 total score is 11.5 points, or 14.2%, according to Tirosh et al. (2008). They determined this score based on a population of 33 youth with motor disabilities aged 4 to 16 years.

Bult et al. (2010) reported that the smallest detectable change based on the confidence interval of 95% for the participation intensity measured with the Dutch CAPE ranges from 1.14 to 1.86 for the five activity types, in youth with and without disabilities (Bult et al., 2010).

For the PedsQL Fatigue and the PedsQL CP no minimal important differences have been reported in the literature. An increase of 25% for one item indicates that the participants reported that problem to occur less. For example, they reported the issue to occur 'never' instead of 'almost never'.

3.6.7 Statistical power

After the study was conducted, statistical power was calculated for the used statistical tests, based on the expected ES, the α-level defined a priori, and the sample sizes as included in the study. G*Power 3, a statistical power analysis programme was used (Faul et al., 2007).

3.6.8 Consideration of the confounding variables

The amount of time youth performed gross motor and upper limb leisure activities, and the amount of physical therapy, as reported by the parents, were each summed for every participant for the study periods between T_1 and T_3 and between T_3 and T_5. Medians of each group were compared for these periods. The participants' weight and height on each measurement occasion of the cross-over design were compared between groups with an independent *t*-test. Additionally, parents were asked to notify the main investigator of any invasive treatments or change in the rehabilitation management occurring over the period of study, as they might impact the assessed outcome variables.

Chapter 4: Results

4.1 Participants

Fifteen children and adolescents with cerebral palsy (CP) participated in the study. The CONSORT diagram (Schulz, Altman, & Moher, 2010) of participant enrolment, stratified and randomised group allocation, and dropout is shown in Figure 4.1. Seven participants were allocated to Group A, receiving the swimming intervention during the first ten weeks, and 8 participants to Group B, the control group during the first ten weeks.

4.1.1 Recruitment

The number of possible candidates assessed for eligibility is unknown. Due to privacy regulations, some therapists and institutions did not reveal the number of eligible participants.

Only two families, approached by the CP reference centre, showed interest in the study; however, one of them did not meet the inclusion criteria. The other candidate was willing to participate. It is unclear how many of the eligible candidates were provided with information. Five children from the consultations in the gait analysis lab showed interest, but only if the swimming programme could be provided closer to their homes. Details of seven families were collected via the specialist in rehabilitation medicine of another hospital, and three of these families agreed to participate.

Ten families of children from the special school were contacted. One of these children had already agreed to participate. One child did not meet the inclusion criteria (due to a planned surgery), and three possible candidates had difficulties to attend, due to a lack of transport. The other five children agreed to participate.

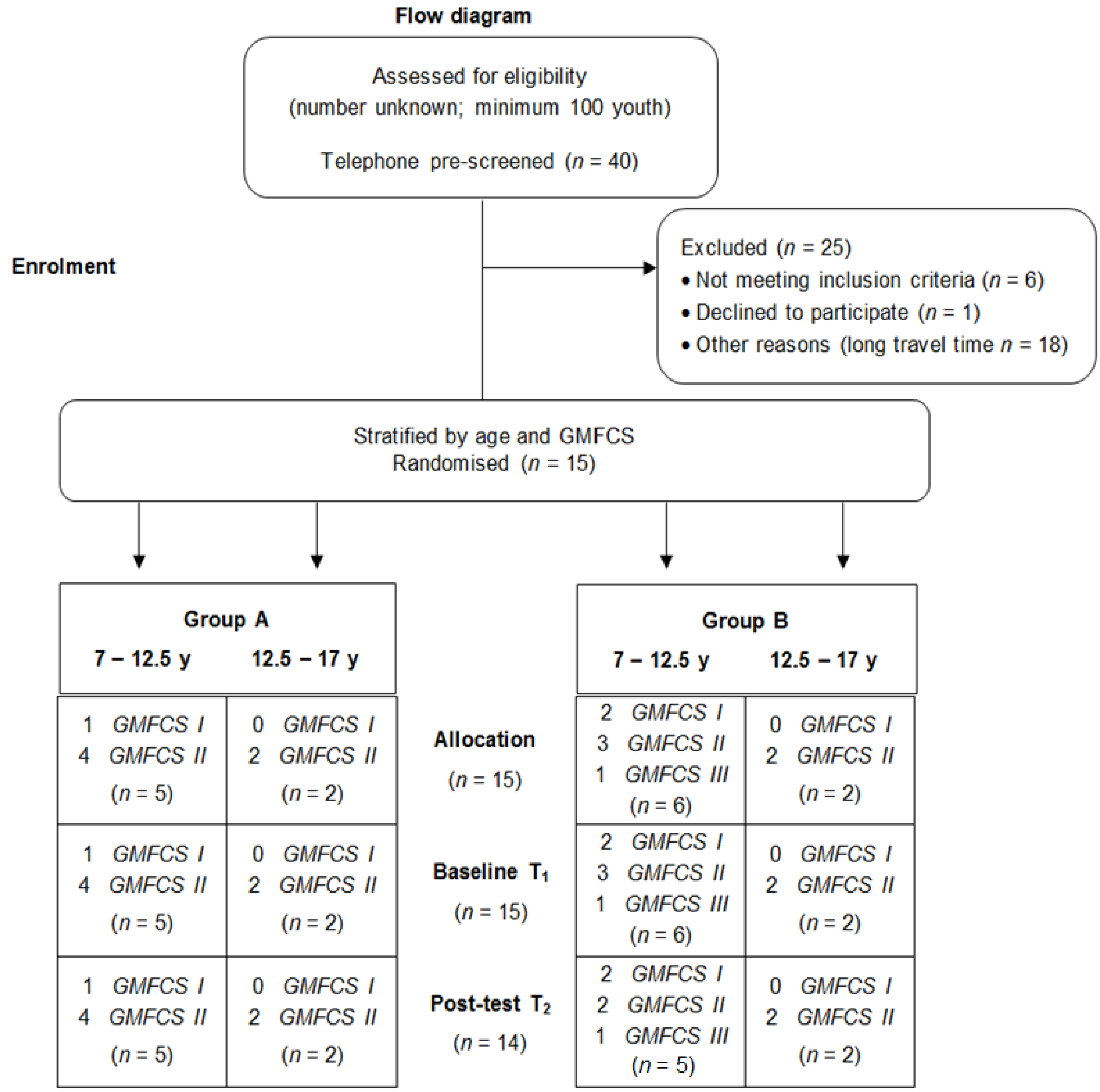


Figure 4.1 Diagram illustrating the flow of participants, including the number for each level of the Gross Motor Function Classification System (GMFCS), within each age category. One child of Group B dropped out at T_2 due to health reasons.

Seven of the 13 private practices that treated youth with CP confirmed repeatedly that no interest was shown by any of the youth or parents that met the inclusion criteria. Fifteen families, informed by the other six therapists, agreed to forward their contact details to the main investigator. Three of these children did not meet the inclusion criteria, and eight of the remaining families did not want to participate if the programme was provided only in Leuven, Belgium. One adolescent showed interest but decided not to participate and the three other children agreed to participate.

Three families of youth that had participated in a previous intervention study conducted by the main investigator agreed to participate. One of these families forwarded the information to another family who agreed to participate as well. The contact details of three other families with a child or adolescent with CP were provided by acquaintances. One of these families was included in the study already, one child did not meet the inclusion criteria and one child did not want to participate if the programme was provided only in Leuven. One parent contacted the main investigator after receiving information from a physical therapist who had obtained the information through an unknown way. This parent's child agreed to participate in the study.

In total, 17 children and adolescents and their families agreed by phone to participate. One month before the study, one of these 17 was excluded due to a medical procedure planned during the study period. The child's sibling withdrew a few weeks later. One month before the start of the intervention, 10 youth of the remaining 15 had confirmed their participation by registering a time slot for the first measurement occasion. One week before the start of the study, all 15 families had confirmed their participation.

4.1.2 Withdrawal

One child from Group B (male, Gross Motor Function Classification System level II, aged 8 years) withdrew before the second measurement (T_2), after the 10-week control period of the cross-over study, due to health problems. Therefore, none of the data for this participant is included in the results.

4.1.3 Participant characteristics

Table 4.1 provides the participant characteristics at baseline. The median age at baseline of the entire sample was 9:2 years and the sample included 8 male and 6 female participants. Most of the participants were classified as Gross Motor Function Classification System level II and both the unilateral spasticity and bilateral spasticity subtypes were most frequent. Six of the participants were born prematurely (< 36 weeks). Most of the youth attended special schools, namely special education types 1, 4 and 8, which are specific to the Flemish special education system, referring to youth with a mild mental, motor, and learning disability, respectively. Both youth attending a mainstream school received *GON*-assistance [‘Integrated education’], i.e. mainstream education with support for youth with special educational needs. The sample included 9 participants with visual impairments such as a cerebral visual impairment or a refractive error.

4.1.4 Comparability at baseline for the descriptive measures

Group A and Group B were similar at baseline regarding demographics, mobility level, manual ability level, gestational age, anthropometrics, type of education, swimming experience, use of orthotic devices and amount of physical therapy (Table 4.1).

Table 4.1 Descriptive information at baseline for the participants of two groups of youth with cerebral palsy (N = 14).

	Group A (n = 7)	Group B (n = 7)	Statistics	p-value
Demographics				
Gender (male/female)	5/2	3/4	Fisher test	.592
Age at enrolment (y:mo)	8:7 (3:4)	11:8 (3:5)	U = 19.0	.535
Age categories			U = 24.5	1.00
7 – 12.5 y	5	5		
12.5 – 17 y	2	2		
Mobility level			U = 24.0	1.00
GMFCS I	1	2		
GMFCS II	6	4		
GMFCS III	0	1		
Manual ability level			U = 24.5	1.00
MACS I	1	1		
MACS II	4	4		
MACS III	2	2		
CP subtype				
Unilateral spasticity	2	3		
Bilateral spasticity	4	2		
Dyskinetic	0	2		
Non – classifiable	1	0		
Gestational age (weeks)	33 (12)	39 (3)	U = 14.5	.220
Anthropometrics				
Height (m)	1.29 (0.3)	1.51 (0.3)	U = 18.5	.478
Weight (kg)	26 (13.0)	35 (16.5)	U = 15.0	.245
Related Medical History				
ASD	2	1		
Speech difficulties	0	2		
Visual impairment	5	4		
Hearing difficulties	0	0		
Seizure disorder	2	2		
Education			Fisher test	.462
Special needs education	7	5		
Mainstream education	0	2		
Swimming experience	6	7	Fisher test	1.00
Orthotic devices	7	6	Fisher test	1.00
Physical therapy (min/week)	90 (60)	100 (45)	U = 24.0	.992

Note. Values are medians (inter quartile range) for the continuous variables and are frequencies for ordinal and categorical variables. GMFCS: Gross Motor Function Classification System; MACS: Manual Ability Classification System; Cerebral palsy (CP) subtypes according to the Surveillance of Cerebral Palsy in Europe; ASD: Autism Spectrum Disorder.

4.2 Statistical power of the study

The recruitment process described in section 3.1.2 resulted in a smaller sample size than required to achieve statistical power of .80 (section 3.6.1). The power of the statistical analyses was calculated after the study was conducted, to assess whether the statistical tests in fact had a fair chance to reject an incorrect null hypothesis.

Power analysis was based on expected effect sizes (ES) of 0.80 for the Water Orientation Test Alyn 2 (WOTA 2) and of 0.50 for all other outcome variables (section 3.6.1). The Mann – Whitney U test for the WOTA 2 had a power of .27, as there were 7 participants in each group. The Mann – Whitney U test for both pain intensity scales had a power of .13, with 6 and 7 participants in Group A and B respectively. The independent *t*-test for the 10-metre walk test (10-MWT), with sample sizes of 7 in each group, had a power of .14. The 1-minute fast walk test (1-min WT), with $n = 6$ and $n = 7$ in the two groups had a power of .13. Only 5 participants were able to complete the PedsQL™ multidimensional fatigue (PedsQL Fatigue) and the PedsQL™ cerebral palsy module version 3.0 (PedsQL CP) questionnaires in Group A, therefore the power for these Mann-Whitney U tests was .12. The Mann-Whitney U tests for the subscales of the Bruininks-Oseretsky test of motor proficiency (2nd edition) and the Pediatric Evaluation of Disability Inventory (PEDI-NL) had a power of .13, with 7 participants in each group. The independent *t*-tests for the Self-Perception Profile for Youth with CP (CBSK-CP) had a power of .13, with 6 and 7 participants in Group A and B respectively. The dependent *t*-tests for the pre-test – post-test design of Research Question 6, questioning the effect of swimming on participation in leisure activities, had a power of .41 with 14 participants included in the sample.

In conclusion, the power of the statistical analyses was low to moderate. Therefore, the probability of making a Type II error (failing to reject a false null hypothesis) was relatively high. In other words, based on statistical significance alone, the chance of incorrectly accepting the null hypothesis was high. Consequently, in this thesis, statistical significance was considered in combination with ES (section 3.6.5) and thresholds for clinical significance reported in the literature (section 3.6.6).

4.3 Compliance with the intervention and perceived exertion

All children and adolescents completed the minimum of 12 sessions of the intervention programme (60%) (section 3.4.1); the participants in Group A attended 16 to 20 sessions and the participants in Group B attended 17 to 19 sessions. Four participants in Group A attended all 20 sessions and the 3 other participants attended 19, 18 or 16 sessions. Three participants in Group B attended 19 sessions and 3 other participants attended 18 sessions. The reasons for not attending a session included heavy snowfall, school field trips, doctor's appointment, youth movement field trips or sickness. The median number of minutes (and inter quartile range in parentheses) the participants spent in the pool during each session was 46 (1.3) min and 48 (7.5) min, ranging from 40 – 50 min and from 40 – 60 min for Group A and Group B, respectively.

The median (inter quartile range) of the ratings of perceived exertion scored by the participants on a scale of 0 to 10 after each swimming session, was 2 (4.5) and 0 (2) for Group A and B respectively. The individual medians ranged from 0 – 10 and from 0 – 6 for Group A and B respectively.

4.4 Consideration of the confounding variables

Three participants were given a botulinum toxin A injection in their lower limbs and a subsequent casting period during their follow-up period (one child in Group A between T_3 and T_4, and 2 adolescents in Group B between T_4 and T_5). One adolescent of Group A underwent brain surgery between T_3 and T_4 due to a ventricular obstruction. One child in Group B had both calves and feet in casts between T_4 and $T_{5,}$ including T_5. One adolescent from Group B had one ankle in a cast between T_4 and T_5, including $T_{5,}$ due to a fracture. Therefore, T_5 was excluded from the analysis of the data from the BA-design of Group B (Figure 3.4), for all outcome variables, since 4 of the 7 participants of Group B had a botulinum toxin A injection and/or a limb in a cast between T_4 and $T_{5.}$ Throughout the study period participants' orthotic devices such as leafsprings and night straighteners were renewed or adapted. One adolescent of Group B had an Osgood-Schlatter knee problem, which was present throughout the study period.

One adolescent of Group A swam at school once every 21 days during the intervention programme. Three participants of Group B swam at school every fortnight throughout the study period. During the follow-up period of Group A (T_3–T_5), 3 participants swam at school less than, or equal to, once every 14 days, and one child went swimming with his father regularly.

During the 4-month period that the Children's Assessment for Participation and Enjoyment (CAPE) questioned at the post-test, one participant in Group B had an orthopaedic surgery for a deformation of one of her feet. Subsequently, she was in a cast, and she was not allowed to undertake any swimming, running or dancing-type activities for 6 months. Another participant of this group changed schools, to a 'boarding school', so his participation in certain activities might have changed due to this alteration of living.

The possible influence of these confounding variables is discussed for each research question in the following chapters.

There was no significant change in the amount of physical therapy, gross motor and upper limb leisure activities over the two 15-week periods for both groups (Table 4.2). There were no statistical differences between groups for height and weight for each measurement occasion (Table 4.3). Nevertheless, Group B was larger and heavier by nearly 7 kg and 8 cm than Group A, at baseline.

Table 4.2 Amount of physical therapy, gross motor function and fine motor function leisure activities (median minutes per week) for all participants (*N* = 14) during the 15-week periods.

	Physical therapy		Gross motor leisure		Fine motor leisure	
Group	T_1–T_3	T_3–T_5	T_1–T_3	T_3–T_5	T_1–T_3	T_3–T_5
	150	150	210	180	60	30
	30	60	142.5	120		
	120	210	240	240	25	25
A	30	15	210	180	60	
	90	90				
	90	75				
	120	150				
Median	90	90	210	180	60	27.5
	120	120	300			
	100	135	210	150	60	
	60	30	180	180	160	100
B	300	270				
	30	30				
	90	90	375	420		
	120	60	360	90		
Median	100	90	300	165	110	100

Note. T_1–T_3 refers to the 15-week period between T_1 and T_3. T_3–T_5 refers to the 15-week period between T_3 and T_5. Not all data were complete.

Table 4.3 Evolution of weight and height over time for all participants (*N* = 14).

Initials	Group	Weight (kg)					Height (m)				
		T_1	T_2	T_3	T_4	T_5	T_1	T_2	T_3	T_4	T_5
JVB	A	26	26	27	28	29	1.29	1.32	1.33	1.345	1.34
FB	A	32	30	32	33	34	1.35	1.35	1.37	1.38	1.37
FT	A	22	22	23	25	25	1.20	1.21	1.23	1.23	1.24
PS	A	40	40	40	41	43	1.62	1.62	1.64	1.65	1.66
EG	A	20	20	21	22	21	1.16	1.18	1.19	1.17	1.18
RR	A	24	27.5	25	26	28	1.24	1.26	1.28	1.29	1.295
FS	A	65	62.5	60	66	65	1.59	1.59	1.60	1.59	1.60
Mean (*SD*)		32.7 (15.8)	32.6 (14.7)	32.6 (13.7)	34.4 (15.3)	35.0 (15.0)	1.35 (0.18)	1.36 (0.18)	1.38 (0.18)	1.38 (0.18)	1.38 (0.18)
AB	B	35	30	33	35	37	1.26	1.27	1.28	1.30	1.35
FP	B	26	25	26	25	26.5	1.27	1.25	1.27	1.28	1.28
LP	B	24	26	27	28	28	1.20	1.21	1.22	1.23	1.24
KR	B	43	45	45	45	–	1.60	1.62	1.65	1.66	–
SD	B	35	35	35	36	–	1.54	1.56	1.55	1.59	–
JS	B	61	57.5	60	60	–	1.62	1.62	1.64	1.64	–
SV	B	51	49	52	52	53	1.51	1.51	1.53	1.55	1.57
Mean (*SD*)		39.3 (13.4)	38.2 (12.5)	39.7 (13.0)	40.1 (12.8)	–	1.43 (0.18)	1.43 (0.18)	1.45 (0.19)	1.46 (0.18)	–
Statistics	*t(12)*	-0.841	-0.774	-1.003	-0.760	–	-0.804	-0.777	-0.746	-0.858	–
	p	.417	.454	.336	.462	–	.437	.452	.470	.407	–

Note. – : no measurement was possible due to casts.

4.5 Comparability at baseline for the outcome variables

The groups were similar at baseline for the WOTA 2 total score ($U = 13.0$; $Z = -1.469$; $p = .165$), for the mental adjustment (MA) subscore ($U = 16.5$; $Z = -1.024$; $p = .335$) and the skills, balance control and movement (SBM) subscore ($U = 15.5$; $Z = -1.158$; $p = .269$). The baseline values of the two groups for the Visual Analogue Scale (VAS), the Faces Pain Scale - Revised (FPS-R), the PedsQL Fatigue, the 10-MWT and the 1-min WT were not significantly different either (VAS $U = 15.5$, $Z = -0.828$, $p = .455$; FPS-R $U = 19.5$; $Z = -0.237$, $p = .857$; PedsQL Fatigue $U = 8.5$; $Z = -1.475$; $p = .154$; 10-MWT $t(12) = -0.317$, $p = .757$; 1-min WT $t(12) = -0.967$, $p = .353$). The groups were comparable at baseline for both bilateral ($U = 22.5$, $Z = -0.261$, $p = .818$) and upper limb ($U = 14.0$, $Z = -1.352$, $p = .191$) coordination and for the self-care ($U = 24.0$, $Z = -0.064$, $p = .973$), mobility ($U = 19.0$, $Z = -0.708$, $p = .508$) and social function ($U = 11.0$, $Z = -1.736$, $p = .090$) subscales of the PEDI-NL. Finally, the groups were comparable at baseline for all subscales of the CBSK-CP and the PedsQL CP (Table 4.4). In summary, there were no significant differences between the groups at baseline for any of the outcome measures.

Table 4.4 Comparison of the groups at baseline for the Self-Perception Profile for youth with cerebral palsy (CBSK-CP) and the PedsQL™ CP module version 3.0 (PedsQL CP).

CBSK-CP			
Subscale	*t(11)*		*p*
Scholastic competence	-1.533		.153
Social acceptance	0.846		.416
Athletic competence	-0.374		.715
Physical appearance	-0.832		.423
Motor competence	-0.160		.876
Global self-worth	-1.396		.190
PedsQL CP			
Subscale	*U*	*Z*	*p*
Daily activities	13.0	-0.736	.510
Social activities	14.5	-0.521	.672
Movement & balance	12.5	-0.868	.438
Pain & hurt	9.5	-1.330	.216
Fatigue	14.0	-0.578	.590
Eating activities	17.0	-0.085	1.00
Speech & communication	16.0	-0.261	.830

Note. The values represent the results of the independent *t*-test and the Mann-Whitney U test.

Chapter 5: Results and discussion of Research Question 1 – Walking ability, pain intensity and feelings of fatigue

5.1 Results

5.1.1 Walking ability

All participants performed both the 10-metre walk test (10-MWT) and the 1-minute fast walk test (1-min WT), with the exception of one adolescent of Group B at T_2. Due to pain he did not perform the 1-min WT.

As shown in Figure 5.1, the baseline scores for the 1-min WT, measuring walking distance at maximum walking speed, were the highest in the participants with Gross Motor Function Classification System (GMFCS) level I that indicates the ability to walk without restrictions, though having limitations in more advanced gross motor skills. Although only 3 participants had a GMFCS level I and only one participant a GMFCS level III, there was a clear relationship between the distances walked during the 1-min WT and the GMFCS level.

Figure 5.2a shows that the change in the 1-min WT scores from baseline (T_1) to post-test (T_2) differed significantly between Group A (n = 7), participating in the swimming intervention (11.6 m increase), and Group B (n = 6), the control group (8.3 m decrease), $t(11) = 2.289$, $p = .043$, CI [0.77 – 39.04], $d = 1.18$, CI [0.00 – 2.37]. No significant differences were observed between groups for the changes over the 15-week period (T_1–T_3) that included a 5-week follow-up period, $t(12) = 1.280$, $p = .225$, CI [-9.83 – 37.83]. However, walking distance in Group A (n = 7) increased 18.9 m compared to a 4.9 m increase in Group B (n = 7) (Figure 5.2b); this difference between groups represents a medium effect size (ES), $d = 0.63$, CI [-0.49 – 1.75]. The walking distance of Group A improved over time (T_1, T_2, T_3); however, the statistical analysis failed to reach significance, $F(2, 12) = 3.788$, MSE = 167.11, $p = .053$. The walking distance in Group B did not change significantly over time, $F(2, 10) = 3.348$, MSE = 119.56, $p = .077$.

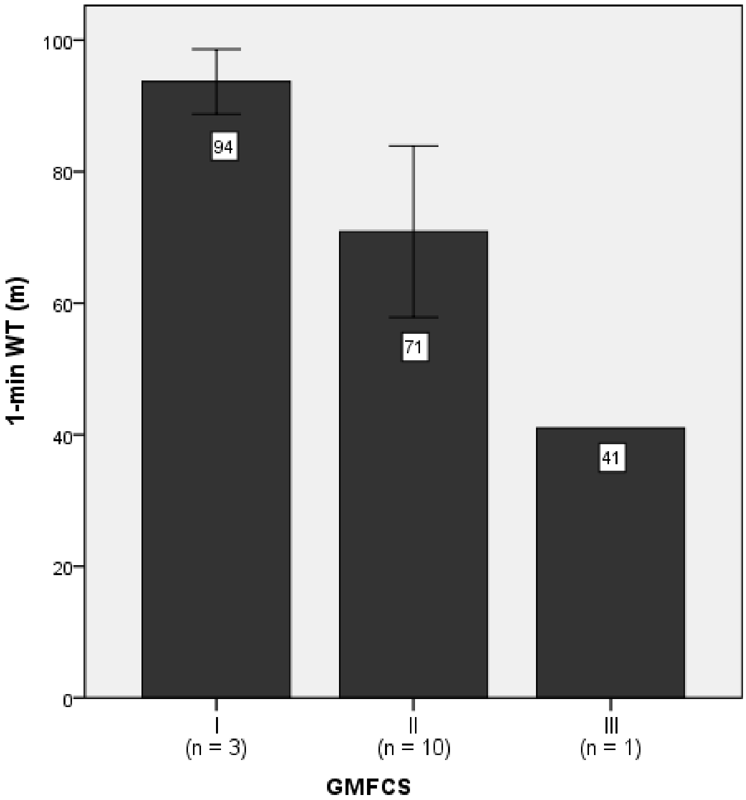


Figure 5.1 Mean baseline values for the 1-minute fast walk test (1-min WT) for each Gross Motor Function Classification System level (GMFCS). Error bars indicate between-subject variability (*SD*).

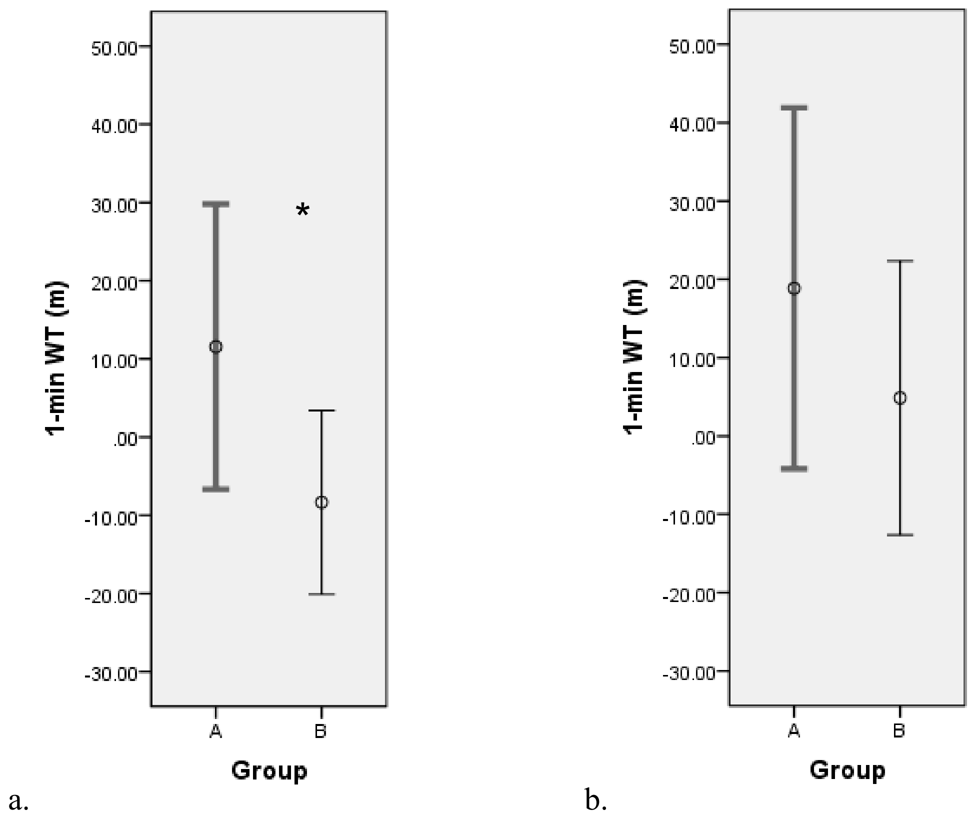


Figure 5.2 The changes over time for the 1-minute fast walk test (1-min WT) for Group A, participating in the swimming intervention, and Group B, the control group; a. The changes over the 10-week intervention/control period; b. The changes over the 10-week intervention/control + 5-week follow-up period. Means +- 1 *SD* are displayed. * *p* < .05.

As the individual results for the 1-min WT in Table 5.1 show, over the 10-week swimming period (T_1–T_2) the distance walked in one minute at maximum speed increased for 5 participants of Group A (a 3.2% to a 63.9% increase relative to baseline). The 2 other participants' walking distance decreased (2.1% and 14.3% relative decrease). Over this 10-week period the walking distance of one child from Group B, the control group, improved (8.2% increase relative to baseline), and one child did not change. The other participants' walking distance decreased ranging from a 4.0% to a 53.7% decrease relative to baseline. Results were different over the 15-week period that included a 5-week follow-up period (T_1–T_3). The distance walked in Group A increased 32% relative to baseline, and the walking distance in all but one of the children increased, ranging from a 1.4% to a 96.3% increase relative to baseline. The mean walking distance in Group B increased 5.6% relative to baseline, over the 15-week control period. Five participants of Group B improved, ranging from a 3.0% to a 46.6% increase relative to baseline. The walking distance in the other 2 participants decreased (by 11.9% and 18.9%), over this 15-week control period.

Table 5.1 Individual and mean results for the 1-minute fast walk test (m).

Initials	Group	T_1	T_2	T_3	T_4	T_5
JVB	A	78	98	96	81	97
FB		62	64	66	64	76
FT		70	60	71	65	75
PS		61	100	105	87	82
EG		96	94	85	62	89
RR		54	84	106	75	81
FS		63	65	87	82	86
Mean (*SD*)		69.1 (14.1)	80.7 (17.4)	88.0 (15.6)	73.7 (10.1)	83.7 (7.7)
AB	B	88	88	129	98	70
FP		41	19	46	62	47
LP		97	105	100	102	100
KR		80	60	84	79	–
SD		74	–	60	65	–
JS		100	96	103	99	–
SV		67	55	59	67	63
Mean (*SD*)		78.1 (20.2)	70.5 (32.1)	83.0 (29.7)	81.7 (17.6)	–

Note. Group A participated in the swimming intervention between T_1 and T_2. Group B participated in the swimming intervention between T_3 and T_4. – : test not performed due to pain or plaster casts.

Analysis of the data from the BA-design of Group B, participating in the swimming intervention following the control period (Figure 3.4b), did not agree with the results of the main analysis of the 1-min WT described above; as changes over their intervention period (T_3–T_4) were not significantly different from the changes over their control period (T_1–T_2), $t(5) = 0.616$, $p = .565$, CI [-19.06 – 31.06]. However, the 1.3 m decrease (3.5% increase relative to T_3) over the swimming period compared to the 8.3 m decrease (15.4% decrease relative to T_1) represents a medium ES, $d = 0.48$, CI [-0.62 – 1.59]. Over this 10-week swimming period the walking distance of 4 participants increased, ranging from a 2.0% to a 34.8% increase relative to baseline

(T_3). The walking distance of 3 participants decreased (ranging from a 3.9% to a 24.0% relative decrease).

After the 20-week follow-up period that followed the completion of the swimming intervention in Group A (T_5), the 1-min WT scores were significantly higher than at baseline, with a mean improvement of 14.6 m since baseline, $t(6) = 3.251$, $p = .017$, CI [3.60 – 25.54], $d = 1.20$, CI [0.06 – 2.33]. The walking distance of all participants but one increased. Although not reaching significance, after the 15-week follow-up period (T_4) walking distance was higher than at baseline (4.6 m mean increase), $t(6) = 0.590$, $p = .577$, CI [-14.40 – 23.55], $d = 0.35$, CI [-0.71 – 1.40]. After this period all but 2 participants' walking distance had increased.

The changes in self-selected walking speed, measured using the 10-MWT, did not differ significantly between Group A, participating in the swimming intervention and Group B, the control group, $t(12) = 0.595$, $p = .563$, CI [-0.39 – 0.69], $d = 0.30$, CI [-0.75 – 1.36] (Figure 5.3a). Table 5.2 shows the individual and group results for the 10-MWT on all measurement occasions. Over the 10-week intervention (T_1–T_2) the mean walking speed of Group A increased 0.02 m/s (or 15.1% relative increase), with the walking speed of 4 participants increasing 10.7% to 72.2% relative to baseline, and the walking speed of 3 participants decreasing 7.2% to 44.9% relative to baseline. Over this 10-week period the mean walking speed of Group B decreased 0.13 m/s (or 7.9% relative decrease), with the walking speed of 3 participants decreasing 26.7% to 37.4% relative to baseline, and the walking speed of 3 participants increasing 2.9% to 18.3% relative to baseline. Similarly, changes over the 15-week period that included a 5-week follow-up period (T_1–T_3), did not differ between groups, $t(12) = 0.340$, $p = .740$, CI [-0.38 – 0.52], $d = 0.17$, CI [-0.88 – 1.22]. The mean self-selected walking speed over the 10 metres decreased in both groups over 15 weeks (Figure 5.3b). Self-selected walking speed did not change significantly over time (T_1, T_2, T_3) for Group A, $F(1.09,6.54) = 0.477$, MSE = 0.18, $p = .530$ (GG) or Group B, $F(2,12) = 1.531$, MSE = 0.05, $p = .256$.

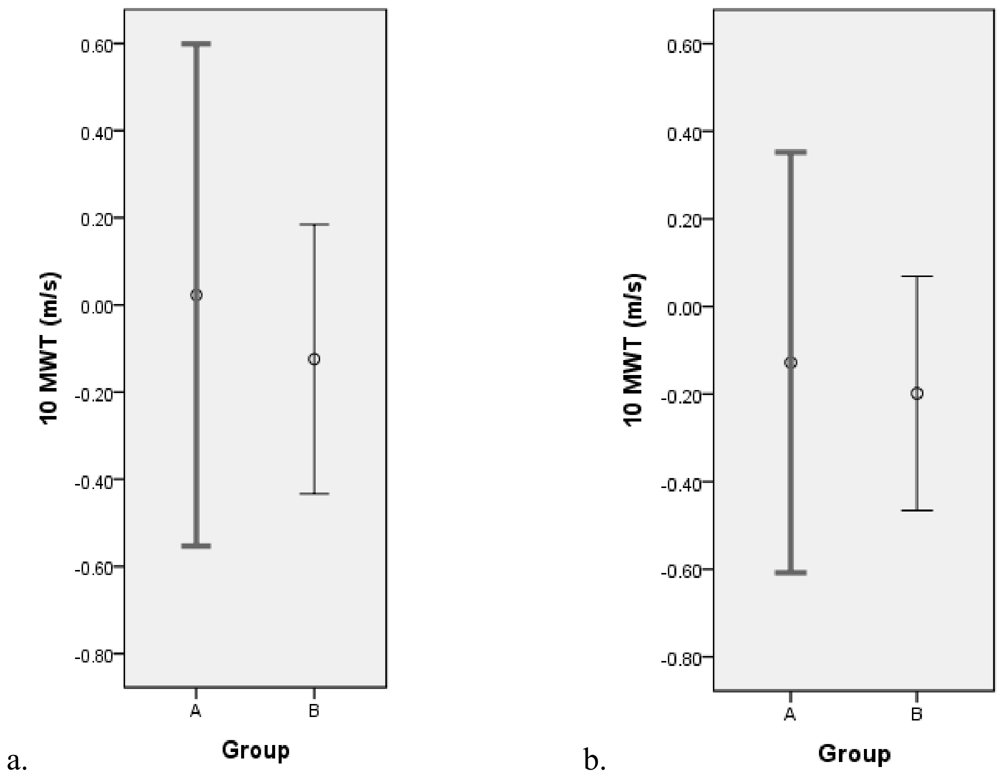


Figure 5.3 The changes over time for the 10-metre walk test (10-MWT) for Group A, participating in the swimming intervention, and Group B, the control group; a. The changes over the 10-week intervention/control period; b. The changes over the 10-week intervention/control + 5-week follow-up period. Means +- 1 *SD* are displayed.

The analysis of the data from the BA-design of Group B agreed with the results of the main analysis of the 10-MWT described above. Changes for the 10-MWT over the intervention period of Group B (T_3–T_4) did not differ significantly from changes over their control period (T_1–T_2), $t(6) = 1.744$, $p = .132$, CI [-0.07 – 0.39]. However, the difference (0.04 m/s increase vs. 0.13 m/s decrease) represents a medium ES, $d = 0.64$, CI [-0.43 -1.72]. Five participants' walking speed improved by 1.1% to 22.1% relative to baseline (T_3). The other 2 participants' walking speed decreased 6.9% and 9.0% relative to baseline (T_3).

Table 5.2 Individual and mean results for the 10-metre walk test (m/s).

Initials	Group	T_1	T_2	T_3	T_4	T_5
JVB	A	0.84	1.21	1.12	0.98	1.35
FB		1.36	1.26	1.18	0.83	1.10
FT		0.83	1.43	1.19	0.93	1.08
PS		0.83	0.92	0.86	0.94	0.84
EG		2.17	1.20	1.17	1.37	1.19
RR		1.02	1.61	1.12	0.83	1.37
FS		1.51	1.10	1.03	1.02	1.02
Mean (*SD*)		1.22 (0.5)	1.25 (0.22)	1.10 (0.12)	0.99 (0.18)	1.14 (0.19)
AB	B	1.43	1.66	0.98	1.20	1.04
FP		1.10	0.69	0.81	0.89	0.95
LP		2.04	1.45	1.67	1.52	1.09
KR		1.25	1.29	1.02	0.95	–
SD		0.87	1.03	0.94	1.08	–
JS		1.22	1.22	1.49	1.52	–
SV		1.17	0.86	0.78	0.78	0.89
Mean (*SD*)		1.30 (0.37)	1.17 (0.34)	1.10 (0.34)	1.13 (0.30)	–

Note. Group A participated in the swimming intervention between T_1 and T_2; Group B participated in the swimming intervention between T_3 and T_4. – : test not performed due to casts.

After the 15- and 20- week follow-up period that followed the completion of the swimming intervention in Group A, the self-selected walking speed for the 10-MWT was not significantly different from baseline. However, the mean self-selected walking speed was lower at the end of the follow-up periods than at baseline. The difference between baseline and 15 weeks of follow-up was -0.24 m/s ($t(6) = -1.672$, $p = .146$, CI [-0.58 – 0.11], $d = -0.59$, CI [-1.66 – 0.48]), and the difference between baseline and 20 weeks of follow-up was -0.09 m/s, $t(6) = -0.438$, $p = .677$, CI [-0.57 – 0.40], $d = -0.22$, CI [-1.27 – 0.83]).

5.1.1.1 **Clinical significance**

Changes larger than 13 m for the 1-min WT were deemed clinically relevant in youth with CP with GMFCS levels I to III (aged 3 to 18 years) (McDowell et al., 2009). In the present study, the mean walking distance on the 1-min WT improved by 18.9 m in Group A over the 15-week period that included the swimming intervention (T_1–T_3). Over the 10-week swimming programmes (T_1–T_2 and T_3–T_4), the walking distance of 3 of the 7 participants in Group A, and one participant in Group B improved by more than 13 m, respectively. Over the 10-week control period of Group B (T_1–T_2) none of the participants' walking distance improved by more than 13 m. After 20 weeks of follow-up (T_5), the walking distance of 5 participants of Group A had improved by more than 13 m compared to baseline (T_1). A change of 0.13 m/s on the 10-MWT represents the smallest detectable change for adults with a spinal cord injury (Lam et al., 2008). In the present study, the self-selected walking speed of 3 participants of Group A, and 2 participants in Group B had improved by more than 0.13 m/s after their respective swimming intervention (T_1–T_2 and T_3–T_4). Over the control period, the walking speed for 2 participants of Group B improved by more than 0.13 m/s.

5.1.2 Pain

One participant of Group A was not capable of completing the pain intensity scales due to limited mental capacity. All participants of Group B completed both scales.

The changes in pain intensity from baseline (T_1) to post-test (T_2) were not significantly different between Group A, participating in the swimming intervention, and Group B, the control group (Table 5.3, Figure 5.4). The scores on the Visual Analogue Scale (VAS) did not change over time (T_1, T_2, T_3) for Group A, $\chi^2(2, n = 6) = 0.333$, $p = .898$ or Group B, $\chi^2(2, n = 7) = 1.778$, $p = .451$. The scores on the Faces Pain Scale - Revised (FPS-R) did not change over time (T_1, T_2, T_3) for Group A, $\chi^2(2, n = 6) = 1.077$, $p = .741$ or Group B, $\chi^2(2, n = 7) = 2.364$, $p = .389$.

Table 5.3 Results of the main statistical analysis comparing the changes over time between Group A (*n* = 6) and Group B (*n* = 7), the swimming and the control group respectively, for the Visual Analogue Scale (VAS) and the Faces Pain Scale–Revised (FPS-R).

Scale	10-week intervention/control period				10-week intervention/control + 5-week follow-up period			
	U	*Z*	*p*	*r*	*U*	*Z*	*p*	*r*
VAS	20.5	-0.075	.991	0.02	17.0	-0.575	.628	0.16
FPS-R	14.0	-1.070	.351	0.30	18.5	-0.408	.788	0.11

Note. The 10-week period represents the change from T_1 to T_2. The 15-week period represents the change from T_1 to T_3. *r* represents the effect size estimate for the Mann–Whitney U test.

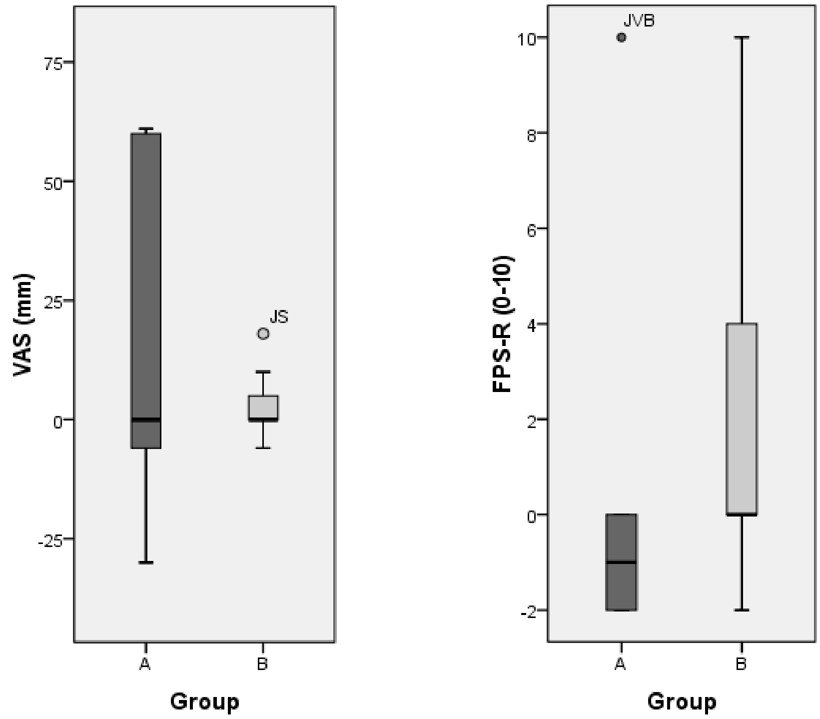


Figure 5.4 The changes over 10-week period (T_1–T_2) for the Visual Analogue Scale (VAS) and the Faces Pain Scale–Revised (FPS-R) for Group A (*n* = 6), participating in the swimming intervention, and Group B (*n* = 7), the control group. Note that participants JS and JVB have outlying scores. In the boxplot of Group A for the FPS-R, the whiskers are close to the quartiles.

There was a high variability for the pain intensity scores within each group and between the two measurement scales; Table 5.4 and Table 5.5 present the individual results for both scales. Between-subject variability for the change in pain intensity between the measurement occasions was high. Some of the participants reported an increase for the pain intensity scale while other participants of the same group reported a decrease for the same pain intensity scale. Additionally, the pain intensity scales showed discrepancies in most of the participants; e.g. an increase for the VAS was reported simultaneously with a decrease for the FPS-R, and vice versa.

Table 5.4 Individual and median results for the Visual Analogue Scale (mm).

Initials	Group	T_1	T_2	T_3	T_4	T_5
JVB		0	61	27	0	32
FB		–	–	–	–	–
FT		30	24	15	0	45
PS	A	30	0	0	0	0
EG		0	0	12	0	0
RR		0	0	0	0	0
FS		13	73	45	17	56
Median (IQR)		6.5 (25.8)	12.0 (51.8)	13.5 (21.0)	0.0 (0.0)	16.0 (41.8)
AB		100	100	0	48	0
FP		0	0	0	0	61
LP		18	28	66	30	5
KR	B	0	0	0	0	0
SD		55	49	47	0	56
JS		0	18	6	6	24
SV		33	33	10	0	21
Median (IQR)		18.0 (44)	28.0 (32)	6.0 (28.5)	0.0 (18)	21.0 (37.5)

Note. Group A participated in the swimming intervention between T_1 and T_2. Group B participated in the swimming intervention between T_3 and T_4. IQR: inter quartile range; – : no response.

Table 5.5 Individual and median results for the Faces Pain Scale - Revised (0 – 10).

Initials	Group	T_1	T_2	T_3	T_4	T_5
JVB		0	10	2	0	4
FB		–	–	–	–	–
FT		2	0	0	0	4
PS	A	0	0	0	0	0
EG		2	0	2	0	0
RR		0	0	0	0	0
FS		2	0	0	0	2
Median (IQR)		1 (2)	0 (0)	0 (1.5)	0 (0)	1 (3.5)
AB		0	10	0	0	0
FP		0	0	0	0	2
LP		2	10	6	0	2
KR	B	0	0	0	0	0
SD		10	10	10	0	6
JS		0	0	0	0	2
SV		4	2	0	0	2
Median (IQR)		0 (3)	2 (10)	0 (3)	0 (0)	2 (1)

Note. Group A participated in the swimming intervention between T_1 and T_2. Group B participated in the swimming intervention between T_3 and T_4. IQR: inter quartile range; – : no response.

Analysis of the data from the BA-design of Group B also revealed the absence of significant changes in the pain intensity scales due to the swimming programme. The changes over time for neither the VAS, $Z = -0.674$, $p = .625$, $r = 0.18$, nor the FPS-R, $Z = -1.473$, $p = .250$, $r = 0.39$, differed significantly between the intervention (T_1–T_2) and the control period (T_3–T_4) of Group B.

After the 15- and 20- week follow-up period that followed the completion of the swimming intervention in Group A, neither the VAS scores differed between baseline and 15 weeks of follow-up (T_4), $Z = -1.089$, $p = .500$, $r = 0.31$, or between baseline and 20 weeks of follow-up (T_5), $Z = -1.095$, $p = .375$, $r = 0.32$, nor the FPS-R scores differed between baseline and T_4, $Z = -1.732$, $p = .250$, $r = 0.50$, or between baseline and T_5, $Z = -0.816$, $p = .750$, $r = 0.24$.

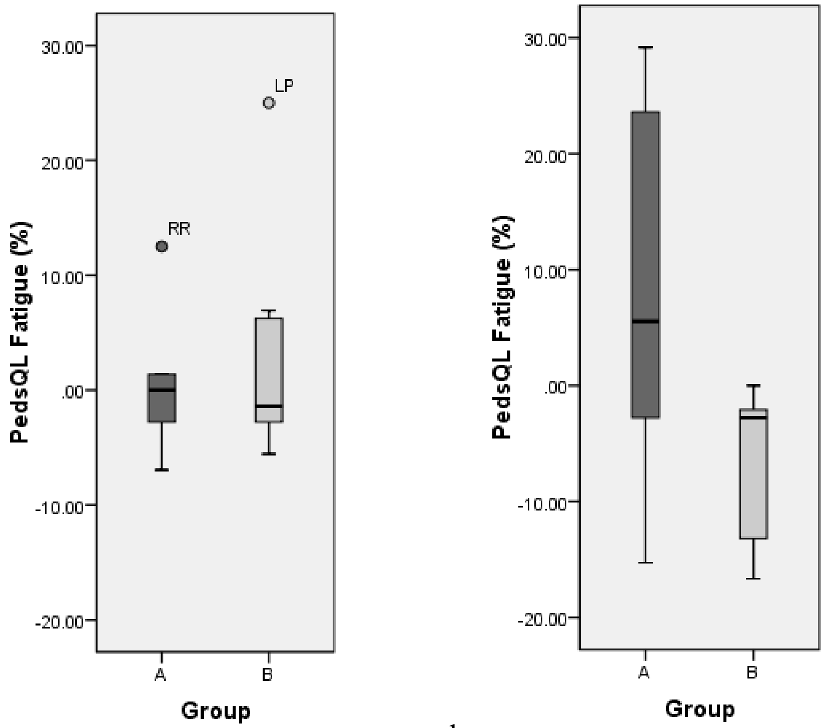
5.1.2.1 **Clinical significance**

Changes larger than 10 mm for the VAS have been reported to represent a significant change in self-reported pain (Stinson et al., 2006). In the present study the median pain intensity score did not increase or decrease more than 10 mm over the swimming period or control period in Group A and B, respectively. Individual results show that 2 participants of Group A, over the swimming intervention, and 2 participants of Group B, over the control period, reported an increase in pain greater than, or equal to 10 mm. In the swimming group (Group A) one participant reported a decrease in pain intensity larger than 20 mm. Von Baeyer (2006) reported that a change of 2 points is the estimate of the minimum clinically significant difference. In the present study the median FPS-R changes over the swimming or control period were not greater than 2 points. Group A included 3 participants whose pain intensity score decreased 2 points over the swimming intervention (T_1–T_2), and one participant's score increased 10 points. Over the control period of Group B (T_1–T_2), the pain intensity scores of 2 participants of Group B increased 8 and 10 points, and one participant reported a decrease of 2 points.

5.1.3 Perceptions of fatigue

Two participants of Group A were not capable of completing the PedsQL^{TM} multidimensional fatigue questionnaire (PedsQL Fatigue) due to limited mental capacity. All participants in Group B completed the scale.

Over the 10-week swimming programme (T_1–T_2), the PedsQL Fatigue scores in Group A (n = 5) did not change, while the PedsQL Fatigue scores in the control group (Group B, n = 7) decreased 1.5% relative to baseline (i.e. an increase in fatigue). The changes over this 10-week period were not significantly different between groups (U = 16.0, Z = -0.245, p = .874, r = 0.07) (Figure 5.5a). Over the 15-week period including 5 weeks of follow-up (T_1–T_3), the PedsQL Fatigue scores of Group A increased 6.2% relative to baseline (i.e. a decrease in fatigue), while the PedsQL Fatigue scores of Group B decreased 2.9% relative to baseline (i.e. an increase in fatigue) (Figure 5.5b). Although the difference between groups was not significant (U = 9.0, Z = -1.390, p = .199), it represents a medium ES (r = 0.40).



a. b.

Figure 5.5 The changes over time for the PedsQL™ multidimensional fatigue Scale (PedsQL Fatigue) for Group A (*n* = 5), participating in the swimming intervention, compared to Group B (*n* = 7), the control group; a. The changes over the 10-week intervention/control period; b. The changes over the 10-week intervention/control + 5-week follow-up period. *Note.* In Figure 5.5a, the whisker is close to the upper quartile in Group A. Participants RR and LP have outlying scores. Note that a positive change in scores reflects a decrease in perceptions of fatigue, while a negative change reflects an increase in perceptions of fatigue.

Table 5.6 shows the individual results for the PedsQL Fatigue. After the swimming intervention of Group A (T_1–T_2), 2 participants of this group reported feeling less fatigued and 2 participants reported feeling more fatigued than at baseline; one participant reported feeling the same level of fatigue. After the 10-week control period of Group B (T_1–T_2), 4 participants reported feeling more fatigued than at baseline and 3 participants reported feeling less fatigued.

Table 5.6 Individual and median results for the PedsQL™ multidimensional fatigue scale (0 – 100).

Initials	Group	T_1	T_2	T_3	T_4	T_5
JVB		68.1	69.4	65.3	84.7	69.4
FB		–	–	–	–	–
FT		–	–	–	–	–
PS	A	88.9	81.9	73.6	84.7	90.3
EG		76.4	73.6	100.0	93.1	90.3
RR		54.2	66.7	83.3	58.3	81.9
FS		90.3	90.3	95.8	94.1	94.4
Median (IQR)		76.4 (20.8)	73.6 (12.5)	83.3 (22.2)	84.7 (8.3)	90.3 (8.3)
AB		94.4	100.0	94.4	98.6	95.8
FP		61.1	58.3	47.2	66.7	84.7
LP		66.7	91.7	54.2	100.0	95.8
KR	B	90.3	97.2	88.9	100.0	100.0
SD		97.2	91.7	94.4	94.4	95.8
JS		94.4	91.7	91.7	77.8	88.9
SV		94.4	93.1	77.8	97.2	88.9
Median (IQR)		94.4 (16.0)	91.7 (3.5)	88.9 (27.1)	97.2 (13.2)	95.8 (6.9)

Note. Group A participated in the swimming intervention between T_1 and T_2. Group B participated in the swimming intervention between T_3 and T_4. IQR: inter quartile range; – : no response. An increase in score reflects a decrease in perceptions of fatigue.

The PedsQL Fatigue scores of Group A (n = 5) did not change over time (T_1, T_2, T_3), $\chi^2(2) = 0.316$, $p = .907$. The PedsQL Fatigue scores of Group B (n = 7) changed significantly over time (T_1, T_2, T_3), $\chi^2(2) = 6.077$, $p = .042$. Post hoc tests revealed a significant increase in fatigue between baseline (T_1) and follow-up (T_3) (Figure 5.6). All participants but one reported feeling more fatigued after this 15-week period that includes 5 weeks of follow-up (T_1–T_3), than at baseline, with decreases in the PedsQL Fatigue scores ranging from a 1.4% to a 16.7% absolute decrease.

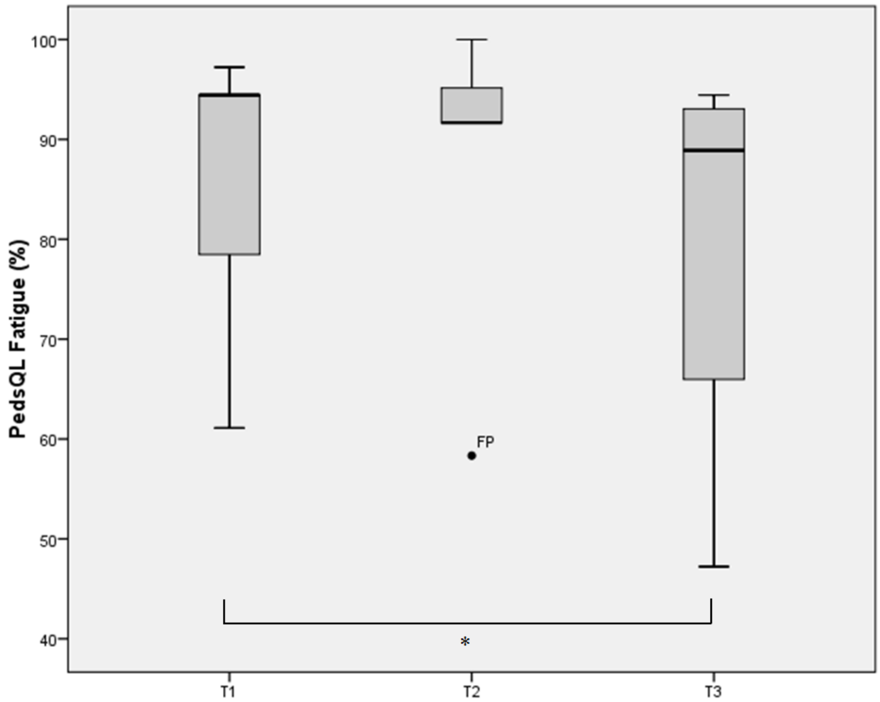


Figure 5.6 Results for the PedsQL™ multidimensional fatigue Scale (PedsQL Fatigue) at T_1, T_2 and T_3 for Group B (n = 7), the control group. ***Note.*** **In the boxplot at T_2 the whisker is close to the quartile. Participant FP has an outlying score. Note that a negative change reflects an increase in perceptions of fatigue. * $p < .05$.**

Analysis of the data from the BA-design of Group B agreed with these results; over the swimming period (T_3–T_4) the PedsQL Fatigue scores increased 12.5% (relative to T_3), while the scores decreased 1.5% over the control period (T_1–T_2) (Figure 5.7). The differences reflect a medium ES, $r = 0.41$, but were not significant, $Z = -1.524$, $p = .141$.

Although not reaching significance, after the 15- and 20-week follow-up period for Group A, the PedsQL Fatigue scores were higher than at baseline. The difference between baseline and 15 weeks of follow-up, 7.7% increase relative to baseline, represents a medium ES ($Z = -1.214$, $p = .313$, $r = 0.38$). The difference between baseline and 20 weeks of follow-up, 4.6% increase relative to baseline, represents a large ES ($Z = -2.023$, $p = .064$, $r = 0.58$), as all participants' PedsQL Fatigue scores had increased since baseline.

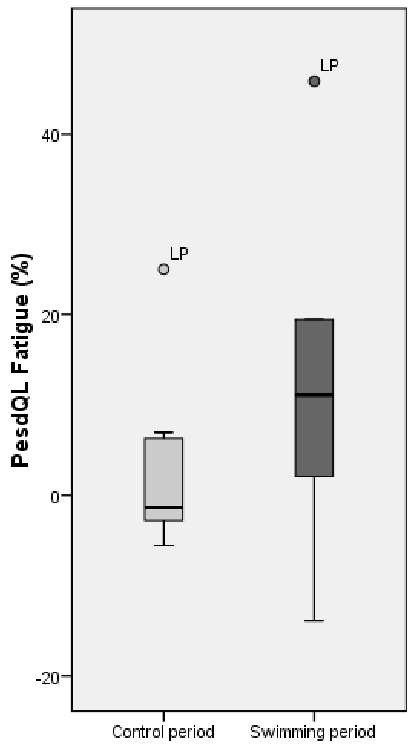


Figure 5.7 Results of the PedsQL ™ multidimensional fatigue scale (PedsQL Fatigue) for changes over the 10-week control period T_1 and T_2 and over the swimming period T_3 and T_4 of Group B (*n* = 7). Participant LP has outlying scores. Note that a positive change in scores reflects a decrease in perceptions of fatigue.

5.1.3.1 **Clinical significance**

For the PedsQL Fatigue, no literature on the minimal important differences was located. An increase of 25% for one item indicates that the participants reported the problem to occur less. For example, they reported the problem to occur ‘never’ instead of ‘almost never’. Median scores of Group A and Group B did not increase or decrease by more than 25%. The PedsQL Fatigue scores of one participant of Group B increased 46% over the swimming intervention (T_3–T_4). ES are reported in the previous section.

5.1.4 Influence of the confounding variables

Renewal of orthotic devices (leafsprings, night straighteners), a knee problem (Osgood-Schlatter) and an ankle fracture could have caused pain during the whole study period and could have affected the walking ability. The participants from Group A that were given a botulinum toxin A injection followed by a period of casting, or underwent a brain surgery, between T_3 and T_4, changed by a similar amount as the median of Group A for the VAS, the FPS-R, and as the mean of Group A for the 1-min WT, over this period. The walking speed for the 10-MWT decreased (21.9%) in the child that was given an injection and increased (10.1%) in the adolescent undergoing surgery, while the mean of Group A decreased (9.1%).

5.2 Discussion

The present study was the first study using a randomised controlled design to investigate the effect of swimming on walking ability, pain intensity and perceptions of fatigue in youth with CP with the ability to walk. Some participants of Group A were not capable of completing the pain intensity scales and the PedsQL Fatigue questionnaire (one and two respectively), which negatively affected the power of the statistical analysis. The findings show that one of the indicators of walking ability, walking distance at maximum walking speed, increased after the 10-week swimming programme, without any adverse effects on pain intensity and perceptions of fatigue. The change in walking distance, measured with the 1-min WT, over the 10-week swimming programme (Group A) was significantly different from the change over this period in the control group (Group B). Twenty weeks after the completion of the swimming programme, a significant increase from baseline was retained. Changes in self-selected walking speed, measured using the 10-MWT, were not significantly different between Group A and Group B. There was a high variability for the changes in pain intensity scores within each group and between the two measurement scales. The changes over time for self-reported pain intensity did not differ significantly between groups. The changes over the control period of Group B for the PedsQL Fatigue scores (increase in fatigue) differed non-significantly from the changes in the scores over the intervention period of Group A (no change) and differed significantly from the changes over the intervention period of Group B (decrease in fatigue), representing small and large ES, respectively. These findings are important with regard to one of the reported causes of pain and fatigue is the physical therapy programme; and with regard to the consequences of increased pain and fatigue on the vicious cycle of deconditioning.

Over the 10-week swimming programme (T_1–T_2), walking distance increased 12 m, in contrast to a decrease of 8 m in the control group. The difference between Group A and Group B was statistically significant and represented a large ES. This was not in agreement with the analysis of the data from the BA-design of Group B, receiving the swimming intervention (T_3–T_4) following the control period (T_1–T_2), though the difference between the intervention and control period of this Group B represented a medium ES. The 1-min WT has not been used in any other aquatic study. However, the improvement of maximum walking speed after an aquatic intervention is in agreement with the literature. The randomised controlled trial of Getz et al. (2012) reported a significant increase in maximum walking speed, measured using the fast walk version of the 10-MWT, after a 4-month adapted swimming programme in a group of young children with a spastic type of CP. Maximum walking speed has not been reported in any other study on aquatics in youth with CP. After a 4-week trunk-targeted exercise intervention, the study of Unger, Jelsma, and Stark (2013) reported a significant 12 m increase on the 1-min WT, which is the same as the increase in the present study. Changes larger than 13 m were deemed clinically relevant (McDowell et al., 2009), which was the case for 3 participants in Group A over the swimming intervention, and for 5 participants in Group A 20 weeks after completing the swimming intervention.

After the control period of Group B (T_1–T_2), the mean distance walked in one minute had decreased 8 m. Smaller changes over the control periods were reported in other intervention studies; Scholtes et al. (2012) and Unger et al. (2013) reported a decrease of 1.2 m and an increase of 1.6 m respectively. In the present study, the walking distance of 2 participants of Group B decreased more than 13 m, the clinically relevant difference, over the 10-week control period. After the 15-week control period these decreases had disappeared. This is reflected in the mean 5 m increase for the 1-min WT over the 15-week control period of Group B (T_1–T_3). These inconsistent changes over the control period might have been due to variations in motivation of the participants.

The walking distance in Group B, participating in the swimming intervention following the control period, improved by only 3.5% compared to a 19.3% improvement in Group A over the swimming period. The inclusion of 2 participants with a dyskinetic type of CP in Group B, compared to the inclusion of only youth with a spastic type of CP in Group A, does not explain the difference in magnitude of change between the groups, because the distance walked in one minute for both dyskinetic participants increased more than the mean increase of Group B, over the swimming period.

In summary, medium to large ES represented the differences between the changes over the swimming intervention (in both Group A and Group B) and the changes over the control period (Group B). The increase in walking distance was retained after 20 weeks of follow-up that followed the completion of the swimming programme in Group A. The scores of Group A differed significantly from baseline, with 5 of the 7 participants of this group increasing by more than 13 m, which was deemed clinically relevant.

Changes in self-selected walking speed over 10 metres did not differ significantly between Group A and Group B, and only a small change over time was present in Group A (0.02 m/s mean increase). This result was in agreement with the within-group comparison of Group B. The effect of an aquatic intervention on walking speed measured with the 10-MWT has been investigated by Getz et al. (2012), but in a much younger sample (3 – 6 year old). Getz et al. (2012) did report improvements (0.11 m/s) after a 4-month adapted aquatics programme for children with CP. The low age range in the sample of the study of Getz et al. (2012) explains their low baseline levels for the 10-MWT, which may have been more likely to improve. However, the studies of Scholtes et al. (2012), Chrysagis, Skordilis, Stavrou, Grammatopoulou, and Koutsouki (2012) and Borggraefe et al. (2010) that included similar age ranges and GMFCS levels as the present study, also reported lower baseline levels (below 1.00 m/s) than the present sample (1.22 m/s and 1.30 m/s for Group A and B respectively). A higher number of participants classified as GMFCS level III than in the present sample ($n = 1$) is a possible explanation.

The increase in maximum walking speed during one minute occurred without an increase in the self-selected walking speed over 10 metres. These findings suggest that youth with CP with high levels of gross motor function are more likely to improve maximum walking speed over a longer distance than self-selected walking speed over a short distance. All participants but one were classified as GMFCS levels I and II. For youth with these levels of gross motor function, walking at a self-selected walking speed over a short distance may not be constrained by their physical limitations to the same extent as walking at a maximum speed for a longer distance. The former task may thus have been completed with close-to-optimal results at baseline, leaving little room for improvement. Thus, for youth with CP with high GMFCS levels a ceiling effect may have been present in the former task. The task of walking at maximum speed for one minute appears more likely to show training effects and thus benefit from the swimming intervention that was offered in the present study. An alteration in the biomechanical constraints during walking in youth with CP due to the decrease of gravity in the water diminishing these constraints might be a possible explanation for this result. However, the biomechanical properties of the gait of the youth with CP were not addressed in this thesis.

To conclude, the present study suggests that walking ability can improve after a 10-week swimming intervention, however only for longer distances at maximal speed. These findings are important as reduced walking speed can limit the ability to keep up with peers, outdoors and in the community (Palisano et al., 2009).

Self-reported feelings of fatigue did not increase after the 10-week swimming intervention. This finding is in agreement with the findings of Kelly et al. (2009), which indicated no changes in fatigue after a 12-week aquatic exercise programme in a sample of five 9 to 11 year-old children with CP. Important to note are the high baseline levels of the sample in the present study (sample median of 89.9%), which indicates that feelings of fatigue occurred rarely in the month prior to baseline. These high scores imply that the youth of the present sample did not feel fatigued frequently, and are similar to scores of typically developing children and adolescents (Gordijn et al., 2011). In the present study, fatigue increased significantly in the control group (Group B) over the 15-week period (T_1–T_3) and this change differed with a medium ES from the change in Group A over this period. Group B subsequently participated in the swimming intervention (T_3–T_4), and changes over this period differed from the changes over the control period with a medium ES as 5 participants of this group reported fewer feelings of fatigue after swimming than at baseline (T_3). Therefore, it can be stated that fatigue did not increase due to the swimming intervention and that the increase in fatigue might be reduced.

Changes over time for the VAS and the FPS-R did not differ between Group A and Group B. The absence of any significant effects was in agreement with the within-group comparison of Group B. The effect of swimming on pain intensity has not been assessed in any other study in youth with CP, so it is impossible to compare these results with others. Nevertheless, in youth with juvenile idiopathic arthritis no change in self-reported pain intensity, measured using the VAS, was reported after a 2-month hydrotherapy (and land-based) programme (Epps et al., 2005). In the present sample the VAS baseline levels were low, and 6 of the 13 participants reported no pain at baseline. Similarly for the FPS-R, baseline levels were low, and 7 of the 13 participants reported no pain at baseline. These findings are in contrast with the results reported by Parkinson et al. (2010) and Parkinson et al. (2013), which indicated that 60% and 74% of the children and adolescents with CP, respectively, report to have pain. The low pain intensity scores for the VAS and the FPS-R reported in the present sample might have been due to actual low pain intensity, or to the non-specificity of the pain the participants were asked to report.

A high variability was present for both pain intensity scales at baseline and throughout the periods of testing. This variability might have been caused by a lack of understanding of the concept of pain or the tool, and/or by a lack of differentiation between accidental and non-accidental pain. The presence of accidental pain might have contributed to the high variability over the control period of Group B, overshadowing the presence of non-accidental pain. Additionally, the pain intensity scales showed discrepancies in most of the participants (e.g. an increase for the VAS simultaneous with a decrease for the FPS-R). This might be explained by the different time range of pain that the scales assess, as the VAS questioned the pain of the past week, and the FPS-R questioned the pain at the moment of testing. Ruskin, Amaria, Warnock, and McGrath (2011) concluded that for a population of youth with chronic pain, not only pain intensity scales but also in depth interviews and questionnaires, questioning the impact of pain on their lives should be used. The PedsQL^{TM} cerebral palsy module version 3.0 (Research Question 3) questions pain in the context of quality of life. The results of this scale are presented in section 7.1 and further discussed in section 7.2.

In conclusion, the findings of the present study suggest that participating in a 10-week swimming programme is likely to improve the maximum walking speed over medium distances (40 – 100 m) of youth with CP (GMFCS I to III), without adverse effects on pain intensity or perceptions of fatigue. The latter is in contrast to regular physical therapy programmes that have been reported causing pain and physical distress (Redmond & Parrish, 2008). Pain and fatigue are perceived to increase due to exercise and consequently are barriers to engaging in physical activity (Verschuren et al., 2012). Additionally, increases in pain and fatigue have been reported to result in locomotion deterioration (Morgan & McGinley, 2013) and to be associated with a higher chance of inactivity (Maltais et al., 2010). Therefore it is pertinent that the engagement in the physical activity programme in the present study did not increase levels of pain or fatigue, and a slight reduction of feelings of fatigue was reported by the participants. As walking with restrictions limits the ability of youth to keep up with peers in the community (Palisano et al., 2009), the improvement in walking speed can facilitate participation in the community. The results of the present study confirm the statement of Murphy and Carbone (2008) that participation in exercise promotes the physical well-being of youth with disabilities. Therefore, swimming can be recommended as a community-based physical activity intervention to combat the cycle of deconditioning.

Limitations of the study and suggestions for future research are discussed in sections 11.3 and 11.4 of the ‘General Discussion’ chapter.

Chapter 6: Results and discussion of Research Question 2 – Upper limb and bilateral coordination, and functional independence

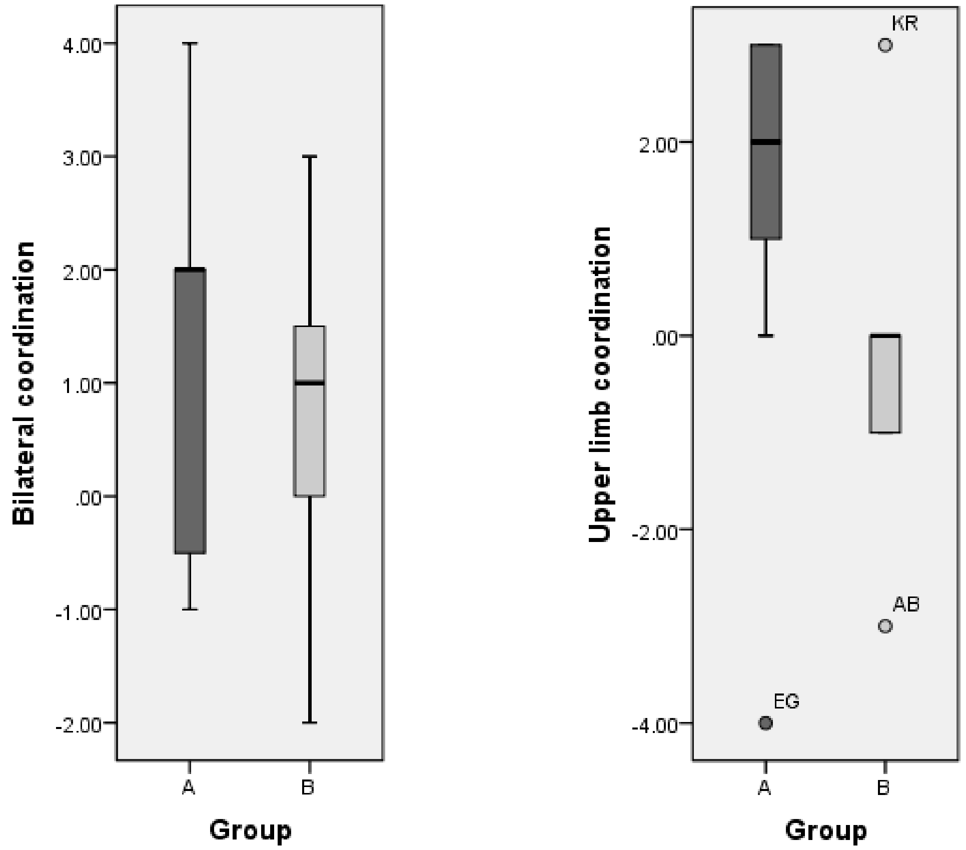
6.1 Results

6.1.1 Coordination

All participants performed the items of the bilateral and upper limb coordination subtests of the Bruininks-Oseretsky test of motor proficiency (2^{nd} edition) (BOT-2).

As shown in Figure 6.1, the change scores of Group A (increase of 2 points for each subtest), participating in the 10-week swimming intervention (T_1–T_2) and the change scores of Group B, the control group (increase of 1 point for bilateral coordination and no change for upper limb coordination), differed without reaching significance, for the bilateral coordination subtest, $U = 21.5$, $Z = -0.390$, $p = .711$, $r = 0.10$, and the upper limb coordination subtest, $U = 13.0$, $Z = -1.506$, $p = .122$, $r = 0.40$. The differences between Group A and B represent small and medium effect sizes (ES), respectively. Over the 15-week period (T_1–T_3) that includes a 5-week follow-up period, the change scores of Group A and B differed with small ES for both bilateral coordination, $U = 18.5$, $Z = -0.799$, $p = .469$, $r = 0.21$, and upper limb coordination, $U = 22.0$, $Z = -0.323$, $p = .775$, $r = 0.09$.

Individual results for the bilateral coordination subtest (Table 6.1) show that the bilateral coordination scores of 4 participants of Group A increased, the scores of one participant remained the same and the scores of 2 participants decreased over the 10-week swimming period (T_1–T_2). Over this 10-week control period in Group B, the bilateral coordination scores of 4 participants increased, the scores of 2 participants remained the same and the scores of one participant decreased. The individual results for upper limb coordination (Table 6.2) show that the scores increased in 5 participants of Group A, remained the same in one participant, and decreased in another participant, over the 10-week swimming period. Over the 10-week control period (Group B), the scores of one participant improved, the scores of 3 participants remained the same and the scores of 3 participants decreased.



a. b.

Figure 6.1 The changes over the 10-week period (T_1–T_2) for the bilateral coordination and upper limb coordination subtests of the Bruininks-Oseretsky test of motor proficiency (2nd ed.) for Group A (n = 7), participating in the swimming intervention, and Group B (n = 7), the control group, are displayed in boxplots. *Note.* In Figure 6.1a the median of Group A is close to the upper quartile. In Figure 6.1b the median of Group B is close to the upper quartile and the whiskers are close to the upper and lower quartiles. Participants EG, AB and KR have outlying scores.

The BOT-2 scores of Group A (n = 7) did not change over time (T_1, T_2, T_3) for bilateral coordination, $\chi^2(2) = 1.238$, $p = .611$, or upper limb coordination, $\chi^2(2) = 3.545$; $p = .189$. Similarly, the BOT-2 scores of Group B (n = 7) did not change over time for bilateral coordination, $\chi^2(2) = 1.182$, $p = .597$, or upper limb coordination, $\chi^2(2) = 2.381$, $p = .346$.

Table 6.1 Individual and median results for subtest 4 of the Bruininks-Oseretsky test of motor proficiency (2nd ed.): bilateral coordination.

Initials	Group	T_1	T_2	T_3	T_4	T_5
JVB		8	10	8	5	7
FB		5	4	5	2	1
FT		8	7	8	6	6
PS	A	8	8	8	7	11
EG		12	14	9	6	8
RR		6	10	5	6	15
FS		5	7	6	6	6
Median (IQR)		8 (2.5)	8 (3)	8 (2.5)	6 (0.5)	7 (3.5)
AB		14	17	8	13	13
FP		4	2	4	3	2
LP		19	21	23	23	23
KR	B	6	7	17	17	–
SD		5	5	4	3	–
JS		6	7	8	11	–
SV		6	6	6	5	5
Median (IQR)		6 (4.5)	7 (6.5)	8 (7.5)	11 (11.0)	–

Note. Group A participated in the swimming intervention between T_1 and T_2. Group B participated in the swimming intervention between T_3 and T_4. Scores are gender- and age-specific. IQR: inter quartile range; – : test not performed due to plaster casts.

Table 6.2 Individual and median results for subtest 7 of the Bruininks-Oseretsky test of motor proficiency (2nd ed.): upper limb coordination.

Initials	Group	T_1	T_2	T_3	T_4	T_5
JVB		4	6	7	4	5
FB		2	5	3	3	5
FT		2	5	5	3	3
PS	A	7	9	7	9	8
EG		6	2	3	1	5
RR		3	6	4	5	8
FS		5	5	5	5	5
Median (IQR)		4 (3.0)	5 (1.0)	5 (2.5)	4 (2.0)	5 (1.5)
AB		9	6	10	7	9
FP		1	1	1	1	1
LP		13	12	19	18	19
KR	B	7	10	19	19	16
SD		4	4	2	4	4
JS		7	7	9	8	7
SV		5	4	4	6	5
Median (IQR)		7 (3.5)	6 (4.5)	9 (11.5)	7 (8.0)	7 (8.0)

Note. Group A participated in the swimming intervention between T_1 and T_2. Group B participated in the swimming intervention between T_3 and T_4. Scores are gender- and age-specific. IQR: inter quartile range.

Analysis of the data from the BA-design of Group B, participating in the swimming intervention following the control period (Figure 3.4b), was partly in agreement with the results of the BOT-2 main analyses described above. There were no significant differences between the changes over the intervention period (T_3–T_4) and the changes over the control period (T_1–T_2) of Group B, for bilateral coordination, Z = -0.084, p = 1.00, r = 0.02, and upper limb coordination, Z = -0.184, p = 1.00, r = 0.05. However, the ES for the difference between the changes over the swimming period and the changes over the control period for upper limb coordination are smaller than the ES representing the difference between Group A and Group B, reported in the main analysis above.

After the 15-week follow-up period (T_4) that followed the completion of the swimming intervention in Group A, the bilateral coordination scores were lower than at baseline (median decrease of 2 points). Although significance was not reached,

Z = -1.897, p = .094, the difference between baseline and T_4 represents a large ES, r = 0.51. Nevertheless, after the 20-week follow-up period (T_5) in Group A, the bilateral coordination scores were not different from baseline, Z = -0.254, p = .891, r = 0.07. Although not reaching significance, the upper limb coordination scores of Group A, after the 15- and 20-week follow-up period, remained higher than at baseline (median increases of 1 point). The differences represent a small ES between baseline and 15 weeks of follow-up, Z = -0.680, p = .688, r = 0.18, and a medium to large ES between baseline and 20 weeks of follow-up, Z = -1.725; p = .156; r = 0.46.

6.1.1.1 **Clinical significance**

Wuang and Su (2009) reported that the minimal important differences (MID) between ‘no change’ and ‘improved’ for the bilateral coordination subtest and the upper limb coordination subtest are 1.1 and 1.6, respectively. In the present study, bilateral coordination scores increased by more than the MID in 4 of the 7 participants of Group A and in 2 of the 7 participants of Group B, over the 10-week swimming programme (T_1–T_2 and T_3–T_4 respectively). Over the control period (T_1–T_2), the bilateral coordination scores of 2 participants of Group B improved by more than the MID. The upper limb coordination scores improved by more than the MID in 5 of the 7 participants of Group A and 2 of the 7 participants of Group B over their respective swimming period. Over the control period of Group B, one participant improved by more than the MID.

6.1.2 Functional independence

The parents of all participants completed the Dutch Pediatric Evaluation of Disability Inventory (PEDI-NL) on all five measurement occasions.

Although not reaching statistical significance, changes over the 10-week period (T_1–T_2) differed between Group A, participating in the swimming intervention and Group B, the control group (Table 6.3). The differences for both the mobility and social function subscales represent medium ES in favour of the swimming intervention. The difference for self-care subscale represents a medium ES in favour of the control period. There were no significant differences between groups for the changes over the 15-week period that includes the 5-week follow-up period (T_1–T_3).

The differences between the Group A and Group B represent small ES for the mobility and self-care subscales and a medium ES for the social function subscale.

Table 6.3 Results of the statistical analyses comparing the changes over time between Group A (*n* = 7) and Group B (*n* = 7), the swimming and the control group respectively, for the Pediatric Evaluation of Disability Inventory.

Subscales	10-week intervention/control period				10-week intervention/control + 5-week follow-up period			
	U	*Z*	*p*	*r*	*U*	*Z*	*p*	*r*
Self-care	11.0	-1.725	.097	0.46	19.0	-0.711	.502	0.19
Mobility	15.5	-1.163	.268	0.31	22.5	-0.257	.829	0.07
Social Function	12.0	-1.602	.123	0.43	14.5	-1.279	.222	0.34

Note. The 10-week period refers to the change from T_1 to T_2. The 15-week period represents the change from T_1 to T_3. *r* represents the effect size estimate for the Mann–Whitney U test.

In Group A, the mobility scores in all participants but one increased or remained the same over the swimming period (T_1–T_2). The mobility scores in Group A improved by a median of 5.3% relative to baseline, compared to no change in the control group (Group B). The parents of all but 2 participants of Group A reported their children to be more capable of performing the items of the social function subscale, after the swimming period than at baseline. The social function subscores in Group A improved by a median of 2.1% relative to baseline, compared to a 1.5% increase relative to baseline in the control group. It can be seen from the data Table 6.4 and Table 6.5 that at baseline, 2 participants of Group B had the maximum score for the mobility subscale and one participant had the maximum score for the social function subscale. The maximum score indicates that the parents reported their children being able to execute all tasks of the PEDI-NL subscale. As the data show in Table 6.6, 6 of the 7 participants of Group A had a lower self-care score than at baseline, after the swimming intervention (mean relative decrease of 2.5%, IQR = 2.5). This decrease is in contrast with changes in Group B over the control period, in which 5 participants increased (mean relative increase of 3.6%, IQR = 6.3).

Table 6.4 Individual and median results for the mobility subscale of the Pediatric Evaluation of Disability Inventory (0 – 100).

Initials	Group	T_1	T_2	T_3	T_4	T_5
JVB		93.7	85.4	83.2	87.6	90.3
FB		83.2	87.6	79.0	83.2	85.4
FT		68.2	83.2	74.7	79.0	76.8
PS	A	81.1	83.2	87.6	87.6	90.3
EG		90.3	90.3	90.3	87.6	90.3
RR		93.7	100.0	100.0	100.0	100.0
FS		66.9	81.1	68.2	62.5	76.8
Median (IQR)		83.2 (17.4)	85.4 (5.8)	83.2 (12.1)	87.6 (6.5)	90.3 (9.2)
AB		100.0	100.0	100.0	100.0	100.0
FP		61.5	54.9	56.8	58.6	60.5
LP		90.3	100.0	100.0	87.6	100.0
KR	B	93.7	93.7	100.0	87.6	100.0
SD		72.8	79.0	74.7	71.1	83.2
JS		100.0	100.0	100.0	100.0	100.0
SV		83.2	79.0	76.8	81.1	83.2
Median (IQR)		90.3 (18.9)	93.7 (21.0)	100.0 (24.3)	87.6 (17.7)	100.0 (16.8)

Note. Group A participated in the swimming intervention between T_1 and T_2. Group B participated in the swimming intervention between T_3 and T_4. IQR: inter quartile range.

Table 6.5 Individual and median results for the social function subscale of the Pediatric Evaluation Disability Inventory (0 – 100).

Initials	Group	T_1	T_2	T_3	T_4	T_5
JVB		70.5	73.8	76.3	71.5	71.5
FB		61.2	60.4	64.7	65.6	64.7
FT		77.7	70.5	73.8	73.8	70.5
PS	A	69.4	70.5	68.5	73.8	70.5
EG		77.7	79.3	79.3	79.3	79.3
RR		61.2	65.6	69.4	71.5	70.5
FS		75.0	81.0	79.3	85.5	83.1
Median (IQR)		70.5 (11.1)	70.5 (8.5)	73.8 (8.9)	73.8 (5.1)	70.5 (4.9)
AB		71.5	72.6	75.0	88.5	76.3
FP		72.6	73.8	77.7	76.3	76.3
LP		81.0	68.5	72.6	83.1	79.3
KR	B	76.3	77.7	79.3	81.0	79.3
SD		77.7	70.5	77.7	77.7	77.7
JS		100.0	92.5	88.5	100.0	88.5
SV		76.3	77.7	72.6	79.3	85.5
Median (IQR)		76.3 (4.9)	73.8 (6.2)	77.7 (4.7)	81.0 (7.3)	79.3 (5.4)

Note. Group A participated in the swimming intervention between T_1 and T_2. Group B participated in the swimming intervention between T_3 and T_4. IQR: inter quartile range.

Table 6.6 Individual and median results for the self-care subscale of the Pediatric Evaluation Disability Inventory (0 – 100).

Initials	Group	T_1	T_2	T_3	T_4	T_5
JVB	A	69.3	73.5	73.5	75.4	76.5
FB		66.3	65.5	68.5	70.9	68.5
FT		75.4	73.5	73.5	76.5	72.6
PS		84.8	80.1	84.8	81.5	80.1
EG		86.9	83.0	93.0	100.0	86.9
RR		74.5	71.7	74.5	75.4	74.5
FS		84.8	83.0	84.8	81.5	84.8
Median (IQR)		75.4 (12.9)	73.5 (9.0)	74.5 (11.3)	76.5 (6.1)	76.5 (8.9)
AB	B	77.6	84.8	84.8	100.0	84.8
FP		57.7	61.3	66.3	63.4	63.4
LP		80.1	83.0	89.5	89.5	86.9
KR		93.0	84.8	93.0	93.0	100.0
SD		69.3	74.5	75.4	70.9	69.3
JS		89.5	89.5	86.9	89.5	93.0
SV		69.3	70.1	67.0	68.5	70.1
Median (IQR)		77.6 (15.5)	83.0 (12.5)	84.8 (17.0)	89.50 (21.6)	84.8 (20.3)

Note. Group A participated in the swimming intervention between T_1 and T_2. Group B participated in the swimming intervention between T_3 and T_4. IQR: inter quartile range.

The self-care subscore of Group A changed significantly over time (T_1, T_2, T_3) (Table 6.7); however, there were no significant differences post hoc. The self-care score decreased the most between T_1 and T_2, with a 1.9 decrease (IQR = 2.15). The mobility and social function subscores did not change significantly over time.

Table 6.7 Results of the statistical analyses of the changes over time (T_1, T_2, T_3) for the Pediatric Evaluation of Disability Inventory.

Group	A (n = 7)		B (n = 7)	
Subscales	$\chi^2(2)$	p	$\chi^2(2)$	p
Self-care	6.870*	.033	2.240	.348
Mobility	3.739	.172	0.111	.981
Social Function	2.296	.358	0.519	.856

Note. Group A participated in the swimming intervention between T_1 and T_2. Group B was the control group. * p < .05.

The analysis of the data from the BA-design of Group B was in agreement with the results of the main analysis of the social function subscale as described above. The social function subscore in Group B changed more over the swimming intervention (T_3–T_4) (9.2% increase relative to baseline) than over the control period (T_1–T_2) (1.5% increase relative to baseline), and significance was reached, $Z = -2.028$, $p = .047$, $r = 0.54$. The change scores of the other subscales did not differ significantly between the swimming and the control period (mobility: $Z = -1.214$, $p = .313$, $r = 0.32$; self-care: $Z = 0.000$, $p = 1.00$, $r = 0.00$). The median mobility and self-care subscores in Group B remained the same over the swimming period.

As the data in Table 6.8 present, after the 15- and 20-week follow-up period (T_4 and T_5 respectively) that followed the completion of the swimming intervention in Group A, the scores on all three subscales were higher than at baseline, although the differences failed to reach significance. The difference between baseline and T_4 for the social function subscore represents a large ES, and the difference between baseline and T_5 for the mobility subscore represents a medium to large ES.

Table 6.8 Results of the statistical analyses of Pediatric Evaluation Disability Inventory scores over a 15- and 20-week follow-up period in Group A (*n* = 7).

Subscales	15-week follow-up period				20-week follow-up period			
	Median difference (IQR)	*Z*	*p*	*r*	Median difference (IQR)	*Z*	*p*	*r*
Self-care	1.1 (6.6)	-1.185	.297	0.32	0.0 (2.5)	0.000	1.00	0.00
Mobility	0.0 (10.0)	-0.943	.438	0.25	6.3 (7.8)	-1.782	.094	0.48
Social Function	4.4 (6.1)	-1.863	.078	0.50	1.6 (4.8)	-1.521	.156	0.41

Note. Median (inter quartile range) differences of the scaled scores (0 – 100) from baseline to 15 weeks follow-up and from baseline to 20 weeks follow-up, and the results of the Wilcoxon signed-rank test are displayed. A positive median difference represents a higher score at follow-up. *r* represents the effect size estimate for the Wilcoxon signed-rank test.

6.1.2.1 **Clinical significance**

The manual of the PEDI (Haley et al., 2005) states that differences larger than twice the standard error, reported in the manual, are clinically significant. The social function scores of 3 participants of Group A improved by more than twice the standard error over the swimming intervention, while the social function scores of 3 participants of Group B decreased (by more than twice the standard error) over the control period. Over the swimming period of Group B, the social function scores of 4 participants improved by more than twice the standard error. The mobility scores of 4 participants in Group A improved by more than twice the standard error over the swimming period, while the mobility scores over the control period of Group B increased for 2 participants and decreased for 2 participants, by more than twice the standard error. Over the swimming period of Group B, the mobility scores of 2 participants increased by more than twice the standard error. The self-care scores of 4 participants of Group A decreased by more than twice the standard error after swimming, while the scores of 4 participants of Group B increased by more than twice the standard error over the control period. Over the swimming period, the self-care scores increased in 2 participants of Group B and decreased in 2 other participants of this group by more than twice the standard error.

6.1.3 Influence of the confounding variables

The child that was given a botulinum toxin A injection between T_3 and T_4 changed by a similar amount as the median of Group A for bilateral coordination and social functioning, over this period. However, the upper limb coordination score decreased by more (2 points) than the median (no change) of Group A in this period, and the self-care and mobility scores (reported by the parents) increased more than the median of Group A. The adolescent of Group A that underwent a brain surgery due to a ventricular obstruction between T_3 and T_4 changed by a similar amount as the median of Group A for bilateral coordination and the mobility subscale, over this period. The self-care score decreased over this period, while the median of Group B increased. The upper limb score and the social functioning score increased more than the median of Group A; however, the difference between T_4 and T_1 was not larger than the median change for this group.

6.2 Discussion

The present study was the first study using a randomised controlled design to investigate the effect of swimming on upper limb and bilateral coordination, and functional independence in youth with CP with the ability to walk. The findings suggest that upper limb coordination and functional independence in mobility and social functioning might improve more over a 10-week swimming intervention than over a control period, in youth with CP. Although statistical significance was reached only in the within-group analysis (BA-design of Group B) for social functioning, the differences between the changes over the 10-week swimming period and the changes over the control period for upper limb coordination and functional independence in mobility represented medium ES. These finding are important as both functional independence and motor proficiency are associated with levels of activity, fitness and participation (Bult et al., 2013; Burns et al., 2009; Kang et al., 2010; Okely, 1999; Tarsuslu & Livanelioglu, 2010; Wrotniak et al., 2006).

Although significance was not reached, upper limb coordination, measured using subtest 7 of the BOT-2, improved more in Group A, participating in the swimming intervention (T_1–T_2) (50% increase relative to baseline), than in Group B, the control group (0% change). The difference between Group A and B represented a medium ES. However, this finding was not in agreement with the within-group comparison of Group B, receiving the swimming intervention (T_3–T_4) following the control period. Bilateral coordination, measured using subtest 4 of the BOT-2, did not change significantly more in Group A over the swimming intervention than in Group B over the control period. Subtests 4 and 7 of the BOT-2 have not been assessed in youth with CP in other aquatic intervention studies, therefore no comparison with previous results could be made. In studies evaluating land-based or other intervention programmes in youth with CP, different subtests of the Bruininks-Oseretsky test of motor proficiency were used. Moreover, these studies mainly reported the use of the first edition of the scale, in contrast to the second edition used in the present study. The first edition can be implemented only until the age of 14.5 years, and was therefore not used in the present study.

The BOT-2 manual (Bruininks & Bruininks, 2005) reports norm reference values (gender- and age- specific) for various paediatric populations such as youth with a developmental coordination disorder (DCD). The baseline values (gender- and age-specific) for bilateral coordination for the present sample were comparable to the norm reference values of the youth with DCD. However, for the upper limb coordination subtest the baseline values were considerably lower than those of the DCD reference sample. Hence, the scores of the present sample can be considered low. A reason for these low baseline levels in the present sample might be the inclusion of youth with a hemiplegic or quadriplegic subtype and youth with visual impairments, which could have affected the eye-hand coordination required during the ball throwing and catching items of this subtest.

Over the swimming intervention (T_1–T_2), the upper limb coordination scores of 5 of the 7 participants of Group A improved by more than the MID, reported for youth with an intellectual disability (Wuang & Su, 2009). In contrast, the upper limb scores in only 2 participants of Group B improved by more than the MID over the swimming intervention (T_3–T_4). The difference between the changes for Group A and Group B over their respective swimming intervention might be explained by the high baseline levels for Group B.

In summary, upper limb coordination might improve after a swimming programme. An improvement in motor coordination is important regarding its effect on the vicious cycle of deconditioning. Learning the skill of swimming, requiring inter-coordination of upper and lower limbs, did not improve bilateral coordination on dry land. Future research should compare the swimming intervention to another physical activity programme to see the additional effects of physical activity in the water.

Despite the known relationship between motor proficiency and functional independence (Öhrvall et al., 2010), the small improvements in upper limb and bilateral coordination are unlikely to have influenced functional independence. Nevertheless, engaging in physical activity in the community may have had an impact on the functional independence of the participants, due to the engagement in activities with peers, with instructors, and other persons present in the swimming pool (social functioning), due to entering and walking around in the pool (mobility), and as youth were encouraged to change and wash themselves independently (self-care).

Social functioning, assessed with the PEDI-NL, improved significantly more over Group B's swimming intervention (T_3–T_4) than over their control period (T_1–T_2). The changes over the 10-week period (T_1–T_2) did not differ significantly between Group A, participating in the swimming intervention, and Group B, the control group; however, the difference between groups represented a medium ES. Additionally, the improvement in social functioning in Group A was maintained 15 and 20 weeks after completing the programme (large and medium ES respectively). Mobility, the second subscale of the PEDI-NL, did not improve significantly more in Group A over the swimming intervention than in Group B over the control period; however, the difference between groups represented a medium ES. The results for the mobility subscale were not in agreement with the within-group comparison of Group B. Changes for the third subscale of the PEDI-NL, self-care, did not differ significantly between Group A, participating in the swimming intervention, and the control group (Group B). In fact, the self-care scores of Group B increased significantly over time.

The improvements for the social functioning and mobility subscale are in agreement with the literature. Aidar et al. (2007) reported a significant increase for social functioning after a 16-week aquatic programme offered to a group of 6 to 13 year-old children with a severe type of CP. The mobility subscale was reported having increased non-significantly after a 14-week aquatic programme in a group of 6 to 11 year-old children with disabilities (including CP) (Fragala-Pinkham et al., 2008), and after a 4-month aquatic programme in 3 to 6 year-old children with CP (Getz et al., 2012). The lack of improvement for the mobility subscale in Group B after their swimming intervention might be due to a ceiling effect, since 4 participants of Group B scored the maximum on the mobility subscale at baseline (T_3). In summary, functional independence in social functioning and mobility might improve after a 10-week swimming programme in ambulatory youth with CP.

The lack of improvement for the self-care subscale after the swimming intervention in the present study is in contrast with the literature, reporting a non-significant increase after a 4-month adapted aquatics programme, and a decrease in self-care scores after a land-based intervention in children with CP (Getz et al., 2012). However, the latter sample included only children aged 3 to 6 years, so their baseline levels were considerably lower than the values of the present sample. Nevertheless, an increase in self-care due to the swimming programme was expected since the participants were required to change and wash themselves, with minimum support provided, before and after each swimming session. Eight of the 15 items of the self-care subscale where practised during these swimming sessions. It was noted by the swimming instructors that assisted the participants to change that some of the participants were able to perform more tasks independently during the final swimming sessions than at the start of the programme. However, 11 of the 14 parents reported no improvements over the 10-week swimming programme, implying that the improvements were not transferred to the tasks performed at home. A longer intervention period may be necessary to show improvements in self-care.

In conclusion, the results of the present study suggest that a swimming programme offered in the community might improve upper limb coordination and functional independence in social functioning and mobility. The present study was the first to report the effects of swimming on coordination in youth with CP. The present study supported the previous findings reporting improvements in social functioning and mobility after an aquatic intervention. The advantages of a swimming programme in the community should be acknowledged since social functioning and mobility are important aspects of participating and engaging in the community, in addition to motor proficiency that influences physical activity, physical fitness and participation levels (Burns et al., 2009; Kang et al., 2010; Wrotniak et al., 2006). Therefore, swimming can be suggested to influence the cycle of deconditioning. The results of the present study confirm the statement of Murphy and Carbone (2008) that participation in exercise promotes the physical and social well-being of youth with disabilities. Limitations of the study and suggestions for future research are discussed in sections 11.3 and 11.4 of the 'General Discussion' chapter.

Chapter 7: Results and discussion of Research Question 3 – Perceived competence, global self-worth and self-reported quality of life

7.1 Results

7.1.1 Self-esteem

One participant of Group A was not capable of completing the Self-Perception Profile for youth with cerebral palsy (CBSK-CP) due to limited mental capacity. All participants of Group B completed the questionnaire. Table 7.1 shows the mean results for the CBSK-CP for both groups for all measurement occasions.

Table 7.1 Mean (*SD*) results for all subscales of the Self-Perception Profile for youth with cerebral palsy.

Group	T_1	T_2	T_3	T_4	T_5
Scholastic competence					
A	3.06 (0.43)	3.23 (0.51)	3.29 (0.60)	3.15 (0.71)	3.33 (0.57)
B	3.45 (0.46)	3.41 (0.64)	3.43 (0.61)	3.34 (0.58)	3.46 (0.69)
Social acceptance					
A	3.08 (0.42)	3.31 (0.39)	3.23 (0.44)	3.40 (0.41)	3.19 (0.49)
B	2.91 (0.32)	3.05 (0.44)	2.95 (0.35)	3.05 (0.61)	2.96 (0.41)
Athletic competence					
A	3.04 (0.79)	2.98 (1.11)	3.27 (0.91)	3.29 (0.93)	3.31 (0.93)
B	3.18 (0.51)	3.04 (0.90)	3.16 (0.77)	3.27 (0.89)	3.21 (0.76)
Physical appearance					
A	3.23 (0.64)	3.48 (0.69)	3.48 (0.69)	3.48 (0.78)	3.56 (0.78)
B	3.46 (0.37)	3.93 (0.10)	3.70 (0.26)	3.59 (0.45)	3.78 (0.41)
Motor competence					
A	2.88 (0.45)	3.29 (0.78)	3.23 (0.66)	3.40 (0.73)	3.35 (0.57)
B	2.93 (0.70)	3.25 (0.80)	3.27 (0.73)	3.45 (0.59)	3.42 (0.60)
Global self-worth					
A	3.23 (0.48)	3.44 (0.45)	3.56 (0.52)	3.63 (0.41)	3.79 (0.40)
B	3.54 (0.30)	3.80 (0.29)	3.66 (0.30)	3.61 (0.51)	3.60 (0.44)

Note. Scores are scaled from 0 to 4, where ‘0’ indicates a low self-perception and ‘4’ indicates a high self-perception. Group A (n = 6) participated in the swimming intervention between T_1 and T_2. Group B (n = 7) participated in the swimming intervention between T_3 and T_4.

As the data in Table 7.2 and Table 7.3 show, the changes over the 10-week (T_1–T_2) and 15-week (T_1–T_3) period were not significantly different between Group A, participating in the swimming intervention, and Group B, the control group.

Table 7.2 Absolute and relative changes over the 10-week intervention/control period for the subscales of the Self-Perception Profile for youth with cerebral palsy for Group A (*n* = 6) and Group B (*n* = 7), the swimming and control group respectively.

Group	Absolute change (/4)	Relative change (%)
Scholastic competence		
A	0.17 (0.45)	6.2 (16.2)
B	-0.04 (0.39)	-1.2 (11.7)
Social acceptance		
A	0.23 (0.60)	9.7 (24.5)
B	0.14 (0.51)	5.8 (18.8)
Athletic competence		
A	-0.06 (0.52)	-4.4 (19.6)
B	-0.14 (0.59)	-5.7 (19.8)
Physical appearance		
A	0.25 (0.35)	8.0 (12.1)
B	0.46 (0.37)	14.5 (13.0)
Motor competence		
A	0.42 (0.50)	13.9 (18.3)
B	0.32 (0.60)	12.7 (26.0)
Global self-worth		
A	0.21 (0.33)	7.1 (11.1)
B	0.27 (0.35)	8.1 (10.3)

Note. Mean (*SD*) values of the changes between baseline (T_1) and post-test (T_2) are displayed. The relative change represents the change over the 10-week period (T_1–T_2) divided by the baseline value, multiplied by 100. Group A participated in the swimming intervention between T_1 and T_2, Group B served as the control group.

After the 10-week swimming intervention, the global self-worth and the perceived competences scores were scored higher than at baseline, except for perceived athletic competence. The perceived motor competence scores increased the most of all subscales over the swimming intervention (increase of 0.42 on a scale of 4, or a 13.9% increase relative to baseline). The scores of 4 participants of Group A improved by 10.3% to 36.4%. Two other participants of Group A reported a 5% lower score than at baseline. The scores for the perceived scholastic competence, social acceptance and motor competence subscales increased more in Group A than in Group B, representing small to medium effect sizes (ES).

Table 7.3 Results of the statistical analyses comparing the changes over time for the Self-Perception Profile for youth with cerebral palsy between Group A (*n* = 6), the swimming group, and Group B (*n* = 7), the control group.

Subscales	10-week intervention/control period				10-week intervention/control + 5-week follow-up period			
	t(11)	*p*	CI	*d*	*t*(11)	*p*	CI	*d*
Scholastic competence	0.878	.405	-0.31 – 0.72	0.47	1.051	.334	-0.33 – 0.82	0.60
Social acceptance	0.276	.788	-0.60 – 0.77	0.15	0.501	.626	-0.38 – 0.60	0.26
Athletic competence	0.253	.805	-0.61 – 0.77	0.13	1.052	.315	-0.27 – 0.77	0.55
Physical appearance	1.079	.304	-0.66 – 0.23	0.54	0.082	.963	-0.45 – 0.48	0.05
Motor competence	0.301	.769	-0.59 – 0.78	0.17	0.045	.965	-0.68 – 0.71	0.02
Global self-worth	-0.315	.759	-0.48 – 0.36	0.16	0.846	.415	-0.33 – 0.75	0.42

Note. The 10-week period refers to the change from T_1 to T_2. The 15-week period represents the change from T_1 to T_3. *d* represents the effect size for the difference between Group A and Group B.

As the results of the statistical analysis in Table 7.4 show, the self-perception scores for both groups did not change significantly over time (T_1, T_2, T_3), except for the physical appearance subscale, which improved significantly in Group B. Contrasts showed a significant change over the 10-week control period (T_1–T_2), $F(1,6) = 11.276$, MSE = 0.13, $p = .015$; the scores of all participants but one improved. However, the change over the 15-week control period (T_1–T_3) was not significant, $F(1,6) = 5.443$, MSE = 0.07, $p = .058$.

Table 7.4 Results of the statistical analyses of the changes over time (T_1, T_2, T_3) for the Self-Perception Profile for youth with cerebral palsy.

	Group A (*n* = 6)			Group B (*n* = 7)		
Subscales	*F*(2,10)	MSE	*p*	*F*(2,12)	MSE	*p*
Scholastic competence	0.695	0.12	.522	0.039	0.06	.962
Social acceptance	0.471	0.17	.638	0.416	0.09	.669
Athletic competence	1.187	0.12	.345	0.352	0.12	.710
Physical appearance	1.750	0.07	.223	7.438**	0.05	.008
Motor competence	2.922	0.10	.100	1.631	0.16	.236
Global self-worth	1.942	0.09	.194	1.404	0.09	.283

Note. Group A participated in the swimming intervention between T_1 and T_2. Group B was the control group. ** $p < .01$.

The analysis of the data from the BA-design of Group B, participating in the swimming intervention following the control period (Figure 3.4b), was partly in agreement with the results of the main analysis for the CBSK-CP described above. Group B's participants' perceived competence scores did not improve significantly more over the swimming period (T_3–T_4) than over the control period (T_1–T_2). The perceived physical appearance scores of Group B improved significantly more over the control period than over the swimming period (Table 7.5). After the 10-week swimming intervention the scores of three subscales had increased, and the scores of the other three subscales had decreased compared to baseline (T_3). The perceived motor competence subscore had increased the most (7.2% increase relative to baseline T_3), which is similar to the results in Group A. However, only the scores of the perceived athletic competence subscale increased more than the changes over the control period (medium ES). This is different from the results of Group A, in which the perceived scholastic competence, social acceptance and motor competence subscores increased more than over the control period.

Table 7.5 Results of the statistical analyses comparing the changes over time for the swimming period (T_3–T_4) and control period (T_1–T_2) of Group B (n = 7) for the Self-Perception Profile for youth with cerebral palsy.

	Mean change (*SD*)		Statistical analysis			
Subscales	(T_1–T_2)	(T_3–T_4)	*t*(6)	*p*	CI	*d*
Scholastic competence	-0.04 (0.39)	-0.09 (0.28)	-0.317	.762	-0.47 – 0.36	0.14
Social acceptance	0.14 (0.51)	0.11 (0.40)	-0.172	.869	-0.57 – 0.49	0.06
Athletic competence	-0.14 (0.59)	0.11 (0.29)	1.015	.349	-0.35 – 0.86	0.50
Physical appearance	0.46 (0.37)	-0.11 (0.58)	-3.833**	.009	-0.94 – -0.21	1.02
Motor competence	0.32 (0.60)	0.18 (0.31)	-0.540	.608	-0.79 – 0.50	0.27
Global self-worth	0.27 (0.35)	-0.05 (0.28)	-1.714	.137	-0.78 – 0.14	0.94

Note. Absolute mean changes of the scores (0 – 4) are presented. *d* represents the effect size for the difference between swimming and control. ** p < .01.

After the 20-week follow-up period that followed the completion of the swimming intervention in Group A (T_5), the perceived motor competence subscore was significantly higher than at baseline (Table 7.6). Four of the 6 participants of Group A reported a higher score after 20 weeks of follow-up than at baseline. After 15 weeks of follow-up (T_4), 5 of the 6 participants of Group A reported a higher perceived motor competence score than at baseline. Although the mean increase of the group failed to reach significance, the difference between baseline and T_4 represents a medium to large ES (Table 7.6). For all other subscales the differences between baseline and T_4 and T_5 represent small to large ES.

Table 7.6 Results of the statistical analyses of the Self-Perception Profile for youth with cerebral palsy (0 – 4) comparing baseline scores and the scores after a 15- and 20-week follow-up period in Group A (*n* = 6).

	15-week follow-up period				20-week follow-up period			
Subscales	Mean difference (*SD*)	*t*(5)	*p*	*d*	Mean difference (*SD*)	*t*(5)	*p*	*d*
Scholastic competence	0.08 (0.68)	0.296	.779	0.14	0.27 (0.41)	1.595	.172	0.49
Social acceptance	0.31 (0.42)	1.831	.127	0.71	0.10 (0.44)	0.581	.586	0.22
Athletic competence	0.25 (0.61)	1.006	.361	0.27	0.27 (0.41)	1.586	.174	0.29
Physical appearance	0.25 (0.64)	0.956	.383	0.32	0.33 (0.49)	1.655	.159	0.43
Motor competence	0.52 (0.54)	2.352	.065	0.79	0.48 (0.44)	2.636*	.046	0.84
Global self-worth	0.40 (0.57)	1.699	.150	0.83	0.56 (0.57)	2.387	.063	1.17

Note. Mean (*SD*) differences from baseline to 15 weeks follow-up and from baseline to 20 weeks follow-up, and the results of the Wilcoxon signed-rank test are displayed. A positive mean difference represents a higher score at follow-up. *d* represents the effect size for the difference between baseline and follow-up.* $p < .05$.

7.1.1.1 Clinical significance

A positive self-perception for all competences and for global self-worth was present at baseline, given the mean scores were larger than 2.5 (Shields et al., 2007). For the perceived motor competence, athletic competence, social acceptance and physical appearance subscales, 4, 3, 2 and one female participants of the total sample, respectively, reported a negative self-perception at baseline. ES are presented in the previous section.

7.1.2 Quality of life

Two participants of Group A were not capable of completing the PedsQL™ cerebral palsy module version 3.0 (PedsQL CP) due to limited mental capacity. All participants of Group B completed the scale.

Table 7.7 shows the medians and inter quartile ranges for all subscale scores throughout the study period. At baseline, all PedsQL CP subscales had a median score of 75.0% or more. This indicates that problems questioned in the scale almost never occurred, representing a high quality of life (QoL). The school functioning, movement and balance and speech and communication subscales had the maximum score of 100% at baseline, in Group A or in Group B. The maximum score indicates that problems with these items never occurred.

The changes over the 10-week (T_1–T_2) and 15-week (T_1–T_3) period were not significantly different between Group A, participating in the swimming intervention, and Group B, the control group, as the results of the statistical analyses presented in Table 7.8 show. The changes over time for all subscales were smaller than 16.7%, and most of the subscores did not change. The scores for the daily activities subscale in Group B increased the most. As the data of Table 7.9 show, none of the subscores of the PedsQL CP changed significantly over time (T_1, T_2, T_3) for Group A or Group B.

Table 7.7 Median (inter quartile range) results for the PedsQL™ CP module version 3.0 (%) for both groups.

Group	T_1	T_2	T_3	T_4	T_5
Daily activities					
A	80.6 (11.1)	91.7 (13.9)	88.9 (19.4)	94.4 (8.3)	94.4 (13.9)
B	77.8 (19.4)	86.1 (13.9)	86.1 (19.4)	88.9 (4.3)	88.9 (23.6)
School activities					
A	93.8 (16.7)	75.0 (25.0)	100.0 (4.7)	100.0 (0.0)	100.0 (4.7)
B	100.0 (9.4)	100.0 (6.3)	100.0 (21.9)	100.0 (25.0)	100.0 (15.6)
Movement & balance					
A	100.0 (5.0)	100.0 (5.0)	95.0 (5.0)	100.0 (0.0)	100.0 (0.0)
B	90.0 (20.0)	100.0 (7.5)	100.0 (10.0)	100.0 (15.0)	100.0 (10.0)
Pain & hurt					
A	87.5 (12.5)	81.3 (6.3)	87.5 (18.8)	81.3 (18.8)	87.5 (12.5)
B	75.0 (28.1)	93.8 (46.9)	93.8 (28.1)	87.5 (15.6)	100.0 (15.6)
Fatigue					
A	81.3 (31.3)	75.0 (25.0)	81.3 (25.0)	87.5 (18.8)	87.5 6.3)
B	75.0 (31.3)	87.5 (25.0)	93.8 (37.5)	100.0 (9.4)	100.0 (9.4)
Eating activities					
A	90.0 (10.0)	85.0 (20.0)	100.0 (10.0)	100.0 (0.0)	100.0 (0.0)
B	95.0 (22.5)	90.0 (17.5)	100.0 (15.0)	100.0 (30.0)	100.0 (12.5)
Speech & communication					
A	100.0 (25.0)	81.3 (25.0)	100.0 (0.0)	100.0 (0.0)	100.0 (0.0)
B	87.5 (18.8)	100.0 (21.9)	100.0 (15.6)	100.0 (15.6)	100.0 (12.5)

Note. Group A (n = 5) participated in the swimming intervention between T_1 and T_2. Group B (n = 7) participated in the swimming intervention between T_3 and T_4. A higher score indicates a higher quality of life.

Table 7.8 Results of the statistical analyses to compare the changes over time between Group A (n = 5) and Group B (n = 7), the swimming and the control group respectively, for the PedsQL™ CP module version 3.0 (%).

Subscales	10-week intervention/control period						10-week intervention/control + 5-week follow-up period					
	Median change (IQR)		U	Z	p	r	Median change (IQR)		U	Z	p	r
Group	A	B					A	B				
Daily activities	2.8 (5.6)	16.7 (18.1)	8.0	-1.548	.139	0.45	0.0 (2.8)	8.3 (19.4)	9.0	-1.405	.191	0.41
School activities	0.0 (8.3)	0.0 (3.1)	14.0	-0.608	.587	0.18	0.0 (16.7)	0.0 (12.5)	10.5	-1.216	.246	0.35
Movement & balance	0.0 (5.0)	0.0 (10.0)	12.0	-0.931	.414	0.27	0.0 (0.0)	0.0 (7.5)	14.0	-0.608	.612	0.18
Pain & hurt	-12.5 (12.5)	-6.3 (15.6)	10.5	-1.151	.299	0.33	0.0 (31.3)	0.0 (12.5)	12.5	-0.828	.444	0.24
Fatigue	-6.3 (12.5)	-6.3 (25.0)	17.0	-0.083	.960	0.02	0.0 (62.5)	-6.25 28.1)	14.5	-0.490	.658	0.14
Eating activities	0.0 (5.0)	0.0 (5.0)	13.0	-0.782	.520	0.23	0.0 (0.0)	0.0 (10.0)	13.0	-0.870	.399	0.25
Speech & communication	0.0 (0.0)	0.0 (12.5)	14.0	-0.609	.597	0.18	0.0 (25.0)	0.0 (12.5)	15.5	-0.347	.777	0.10

Note. The 10-week period refers to the change from T_1 to T_2. The 15-week period represents the change from T_1 to T_3. IQR: inter quartile range. r represents the effect size estimate for the Mann–Whitney U test.

Table 7.9 Results of the statistical analysis of the changes over time (T_1, T_2, T_3) for the PedsQL™ CP module version 3.0.

	Group A (n = 5)		Group B (n = 7)	
Subscales	$\chi^2(2)$	p	$\chi^2(2)$	p
Daily activities	1.778	.478	3.900	.175
School activities	3.818	.222	0.143	.981
Movement & balance	0.200	1.00	1.733	.528
Pain & hurt	2.842	.270	2.381	.346
Fatigue	0.353	.920	1.826	.442
Eating activities	3.714	.333	2.375	.309
Speech & communication	3.800	.333	1.733	.528

Note. Group A participated in the swimming intervention between T_1 and T_2. Group B was the control group.

Analysis of the data from the BA-design of Group B also revealed the absence of significant differences in the PedsQL CP subscores between swimming and no swimming, except for the daily activities subscale. The scores of Group B for this subscale increased significantly less over the intervention period (T_3–T_4) than over the control period (T_1–T_2), with a 1.0 % absolute increase compared to 16.7% absolute increase, as shown in Table 7.10. The scores of 4 participants improved over the intervention period and the scores of 5 participants improved over the control period. Similar to the changes in Group A, most of the subscales did not change over the swimming period.

Table 7.10 Results of the statistical analyses comparing the changes over the swimming period (T_3–T_4) and control period (T_1–T_2) of Group B (*n* = 7) for the PedsQL™ CP module 3.0 (%).

	Median change (IQR)		Statistical analysis		
Subscales	(T_1–T_2)	(T_3–T_4)	*Z*	*p*	*r*
Daily activities	16.7 (18.1)	1.0 (11.1)	-2.028*	.047	0.54
School activities	0.0 (0.1)	0.0 (0.1)	-0.816	.750	0.22
Movement & balance	0.0 (10.0)	0.0 (2.5)	-0.730	.625	0.20
Pain & hurt	-6.3 (15.6)	0.0 (21.9)	-0.813	.500	0.22
Fatigue	-6.3 (25.0)	6.3 (37.5)	-1.265	.281	0.34
Eating activities	0.0 (5.0)	0.0 (12.5)	-1.095	.375	0.29
Speech & communication	0.0 (12.5)	0.0 (0.1)	-0.962	.500	0.26

Note. Absolute median change scores are presented. *r* represents the effect size estimate for the Wilcoxon signed-rank test. IQR: inter quartile range.
* $p < .05$.

After the 15- and 20-week follow-up period that followed the completion of the swimming intervention in Group A (*n* = 5), the PedsQL CP subscores did not differ significantly from baseline. As the data in Table 7.11 show, the differences between baseline and follow-up represent small to large ES. The fatigue subscale had the largest absolute increase, but a high variability within the group was present.

Table 7.11 Results of the statistical analyses of the PedsQL™ CP module 3.0 (%) comparing baseline scores and the scores after a 15- and 20-week follow-up period in Group A (*n* = 5).

	15-week follow-up period				20-week follow-up period			
Subscales	Median difference (IQR)	*Z*	*p*	*r*	Median difference (IQR)	*Z*	*p*	*r*
Daily activities	5.6 (2.8)	-1.219	.313	0.39	2.8 (5.6)	-1.604	.250	0.51
School activities	6.3 (16.7)	-1.604	.250	0.51	6.3 (16.7)	-1.289	.375	0.41
Movement & balance	0.0 (0.0)	-0.447	1.00	0.13	0.0 (5.0)	0.000	1.00	0.00
Pain & hurt	-6.3 (6.3)	-0.736	.625	0.23	0.0 (18.8)	-0.552	.750	0.17
Fatigue	12.5 (37.5)	-1.095	.375	0.35	12.5 (31.3)	-1.604	.250	0.51
Eating activities	0.0 (10.0)	-1.414	.500	0.45	10.0 (10.0)	-1.473	.250	0.47
Speech & communication	0.0 (12.5)	-1.342	.500	0.42	0.0 (25.0)	-1.342	.500	0.42

Note. The 15-week follow-up period represents the change between T_1 and T_4. The 20-week follow-up period represents the change between T_1 and T_5. A positive median difference represents a higher score at follow-up. IQR: inter quartile range. *r* represents the effect size estimate for the Wilcoxon signed-rank test.

7.1.2.1 Clinical significance

For the PedsQL CP, no literature on the minimal important differences was located. An increase of 25% for one item indicates that the participants reported the problem to occur less. For example, they reported the problem to occur ‘never’ instead of ‘almost never’. Median scores of Group A and Group B did not increase or decrease by more than 25%. ES are reported in the previous section.

7.1.3 Influence of the confounding variables

The motor competence subscore of the participant that was given a botulinum toxin A injection and of the participant that underwent brain surgery did not change between T_3 and T_4, compared to a small increase for the median of Group A.

7.2 Discussion

The present study was the first study using a randomised controlled design to investigate the effect of swimming on self-perception and QoL in youth with CP. Some participants of Group A were not capable of completing the CBSK-CP and the PedsQL CP questionnaires (one and two respectively), which negatively affected the power of the statistical analysis. Over the swimming intervention, the perceived motor competence scores increased the most of all subscales, improving in 4 of the 6 participants; however no significant difference with the control group was present. Perceived motor competence is the subscale, added in the Dutch version of Harter’s self-perception profile for children/adolescents adapted for youth with CP (Komdeur et al., 2001). The items in this subscale question perceived competence in running, ball skills, jumping, swimming and cycling. Since perceived sports competence is reported to act as a mediator between childhood motor skill proficiency and adolescent physical activity and fitness levels in the general population (Barnett et al., 2008), an improvement is of importance with regard to the cycle of deconditioning. Other effects of swimming on perceived competence, global self-worth and QoL were absent. A longer intervention period might have led to (larger) changes in self-perception and QoL.

All perceived competence scores and the global self-worth score of Group A improved over the 10-week swimming intervention (T_1–T_2), except for perceived athletic competence. The perceived motor competence subscore in Group A improved by a mean of 0.42 (possible score range 0 – 4), and improved in 4 of the 6 participants in this group. This increase in perceived motor competence was retained 15 and 20 weeks after the completion of the swimming programme. A similar trend towards improvement for perceived competence was reported by Thorpe et al. (2005) using a case series design including 7 youth with CP, which indicated improvements in all perceived competences, except for global self-worth, after an aquatic exercise programme. No values were reported in this study, so no comparison could be made. In Group B, the perceived social acceptance, physical appearance, motor competence and global self-worth scores improved over the 10-week control period. These results suggest that the trend towards improvement following the swimming intervention in Group A, cannot be attributed solely to the aquatic intervention.

The general improvement in Group A over the intervention period was not significantly different from the changes in Group B over the control period. However, Group A reported larger improvements for the perceived motor competence, scholastic competence and social acceptance subscales than the control group (Group B), with differences between groups representing small to medium ES. In comparison with the randomised controlled trial reported by Verschuren et al. (2007), the perceived athletic competence subscore of the exercise group increased significantly more (11%) than in the control group after an 8-month land-based exercise programme. In the present study the mean subscore of perceived motor competence was the only subscore improving by more than 10% over the 10-week swimming programme in Group A. In the present study, scores in neither group changed significantly over time, except for the perceived physical appearance subscale that increased significantly over the 10-week control period in Group B. However, there was no significant increase over the 15-week control period (T_1–T_3), suggesting that this change was not clinically relevant.

The perceived motor competence and social acceptance subscales had the lowest baseline values. In general, baseline values for both groups indicated a positive self-perception, similar to previously reported values for youth with CP (Komdeur et al., 2001; Schuengel et al., 2006; Shields et al., 2007). At baseline, 4, 3, 2 and one participants of the total sample reported a negative self-perception for the perceived motor competence, athletic competence, social acceptance and physical appearance subscales, respectively. Interestingly these participants were all female. In the studies of Shields et al. (2007) and Schuengel et al. (2006) the female CP subsample reported a negative self-perception for athletic competence, but not for the other subscales.

Regarding the QoL questionnaire, the PedsQL CP, none of the subscale scores changed significantly due to the swimming programme. This finding contradicts Groff et al. (2009) who reported that participation in adapted sports influences QoL. The effects of an aquatic programme on QoL have not been investigated in any other study, so no comparison to other aquatic programmes could be made.

In studies that assess QoL with the PedsQL CP in youth with CP (Redman, Finn, Bremner, & Valentine, 2008; Tantilipikorn, Watter, & Prasertsukdee, 2013; Varni et al., 2006), the baseline values for all subscales were lower than the baseline values of the present sample, except for the subscale pain and hurt (Tantilipikorn et al., 2013). This difference in baseline values might be explained by the inclusion of all Gross Motor Function Classification System levels in the two samples of Tantilipikorn et al. (2013) and Varni et al. (2006), compared to only the levels I, II and III in the current sample. The trend towards improvement for the subscale daily activities over the control period was also reported by Redman et al. (2008), who showed a significant linear trend for the daily activities and the pain and hurt scores during a 6-month control period.

There were no changes in QoL over the 10-week intervention programmes in Group A and B. This absence in change might be due to the baseline values of both groups being higher than 75%, which indicates that the problems questioned in the scale almost never occurred. Additionally, since QoL is affected by a variety of factors, the duration of the intervention may have been too short to create a significant change. However, since increases in QoL have been reported after 12-week exercise programmes (Demuth et al., 2012; Engsberg, Ross, & Collins, 2006; Gates et al., 2012), 10 weeks was deemed long enough to affect QoL, at the start of the study. Further investigation using a longer intervention period is required to fully assess the effect of the swimming intervention on QoL.

The subscore of the pain and hurt subscale did not change over time or between groups, which is in agreement with the results of the Visual Analogue Scale and the Faces Pain Scale-revised presented in Chapter 5. This similar result confirms that the swimming intervention did not increase pain.

To conclude, the present study was the first randomised controlled trial to investigate the effect of a swimming intervention on self-perception and QoL in youth with CP. There was some indication that perceived motor competence might improve after a 10-week swimming programme, and that improvements can be retained for a minimum of 20 weeks after completing the programme. This possible improvement important since perceived motor competence mediates the association between childhood motor skill proficiency, and adolescent physical activity and fitness levels (Barnett et al., 2008). The relatively high baseline levels for self-perception and QoL, in addition to the short period over which these constructs were assessed, might have contributed to the absence of changes for the other self-perception scales and QoL.

Limitations of the study and suggestions for future research are discussed in sections 11.3 and 11.4 of the ‘General Discussion’ chapter.

Chapter 8: Results and discussion of Research Question 4 – Mental adjustment and swimming skills

8.1 Results

8.1.1 Mental adjustment and motor skills in the water

All participants performed the items of the Water Orientation Test Alyn 2 (WOTA 2), assessing mental adjustment and motor skills in the water.

Since the sample was relatively small and variability among participants was high, the individual results for the total score of the WOTA 2 are shown in Table 8.1. As the data in Table 8.2 present, the WOTA 2 scores increased significantly more in Group A, participating in the swimming intervention (33.3% absolute increase) than in Group B, the control group (6.2% absolute increase). This effect was considered large, $r = 0.77$. The increases of the total score in Group A ranged from a 12.4% to a 38.7% absolute increase. Changes were significantly different between groups for the mental adjustment (MA) and the skills, balance control and movement (SBM) subscale, with large effect sizes (ES) for both. Over the 15-week period (T_1–T_3) that includes a 5-week follow-up period, the improvement on the WOTA 2 total score was still present, with a total median absolute increase of 34.6% for Group A, which was significantly different from the 3.7% absolute increase for Group B. Also the changes in MA and SBM subscores over the 15-week period differed significantly between groups (Table 8.2).

Table 8.1 Individual and group results for the total score of the Water Orientation Test Alyn 2 (WOTA 2).

WOTA 2 (%)	Group	T_1	T_2	T_3	T_4	T_5
JVB	A	40.0	74.7	74.7	82.7	81.3
FB	A	13.3	38.7	38.7	36.0	29.3
FT	A	36.0	65.3	65.3	56.0	56.0
PS	A	46.9	80.2	81.5	86.4	80.3
EG	A	41.3	80.0	84.0	84.0	86.7
RR	A	38.7	76.0	77.3	80.0	81.3
FS	A	44.4	56.8	56.8	55.6	56.8
Median (IQR)		40.0 (5.6)	74.7 (16.9)	74.7 (18.3)	80.0 (27.6)	80.3 (24.9)
AB	B	52.0	69.3	64.0	97.3	97.3
FP	B	16.0	14.7	13.3	33.3	33.3
LP	B	49.3	73.3	78.7	94.7	96.0
KR	B	53.1	64.2	56.8	86.4	–
SD	B	4.9	9.9	11.1	45.7	–
JS	B	87.7	92.6	86.4	95.1	–
SV	B	59.3	65.4	60.5	90.1	88.9
Median (IQR)		52.0 (23.5)	65.4 (31.9)	60.5 (36.3)	90.1 (28.8)	–

Note. Group A participated in the swimming intervention between T_1 and T_2. Group B participated in the swimming intervention between T_3 and T_4. IQR: inter quartile range; – : test not performed due to plaster casts.

Table 8.2 Results of the statistical analyses to compare the changes over time between Group A (n = 7) and Group B (n = 7), the swimming and the control group respectively, for the Water Orientation Test Alyn 2 (%).

Period	Scale	Median change (IQR)		Statistical analysis			
		Group A	Group B	U	Z	p	r
10-week intervention/ control period	TOT	33.3 (8.7)	6.2 (9.3)	2.0**	-2.878	.002	0.77
	MA	24.2 (4.6)	0.0 (9.2)	0.0**	-3.148	.001	0.84
	SBM	35.7 (17.9)	16.7 (21.4)	8.5*	-2.056	.042	0.55
10-week intervention/ control + 5-week follow-up period	TOT	34.6 (9.3)	3.7 (9.1)	2.5**	-2.814	.003	0.75
	MA	30.3 (3.3)	-2.6 (13.3)	1.0**	-3.006	.001	0.80
	SBM	38.1 (15.5)	4.8 (20.2)	6.0*	-2.380	.016	0.64

Note. The values represent the median absolute changes and inter quartile ranges (IQR). TOT = total score; MA = mental adjustment subscore; SBM = skills, balance control and movement subscore. * p < .05; ** p < .01. r represents the effect size estimate for the Mann–Whitney U test.

The total score, the MA subscore and the SBM subscore of Group A (n = 7) changed significantly over time (T_1, T_2, T_3), as the statistical results show in Figure 8.1 and Figure 8.2. The figures also present the results of the post hoc tests, revealing significant differences between baseline (T_1) and post-intervention (T_2) and between baseline and the end of the 5-week follow-up period (T_3) for the total score and both subscores. As can be seen in Figure 8.2, the SBM subscore changed significantly over time for Group B, the control group, and post hoc tests revealed a significant difference between baseline (T_1) and post-test (T_2) (16.7% absolute increase); however, this increase was not retained after a further 5 weeks of follow-up (4.8% absolute increase since baseline).

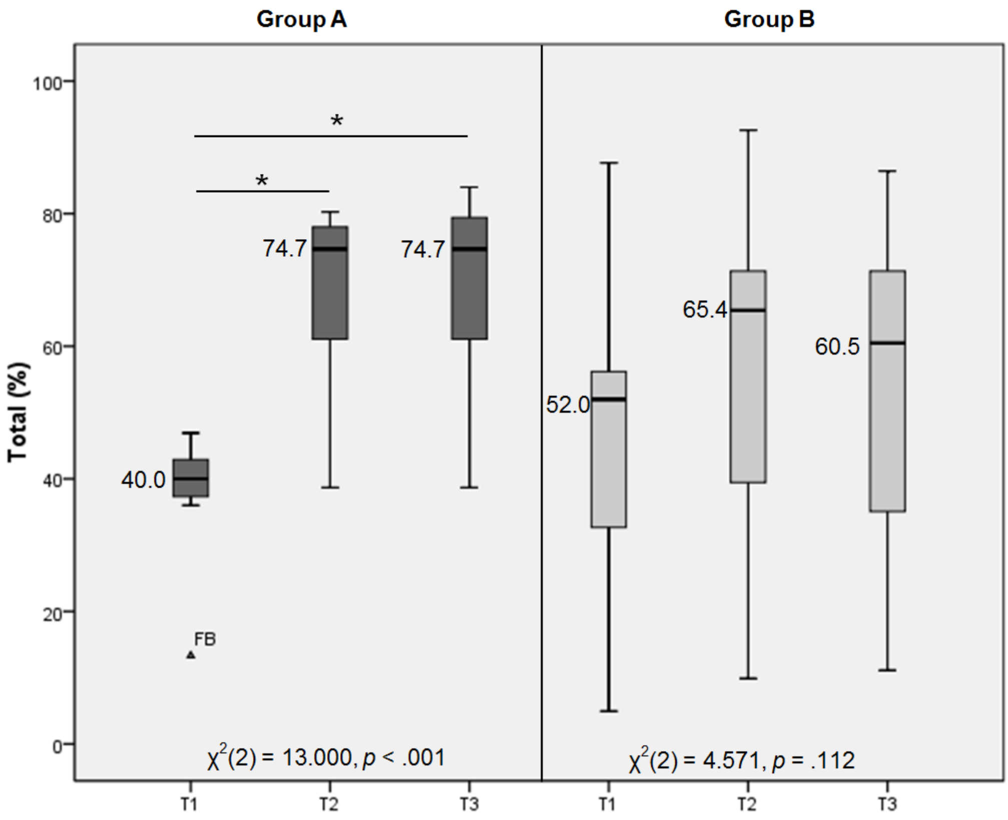


Figure 8.1 The total score of the Water Orientation Test Alyn 2 at T_1, T_2, T_3 and the results of the statistical analysis of the changes over time (T_1, T_2, T_3). Group A (*n* = 7) participated in the swimming intervention between T_1 and T_2. Group B (*n* = 7) was the control group. Note that Participant FB has an outlying score at baseline. * *p* < .05.

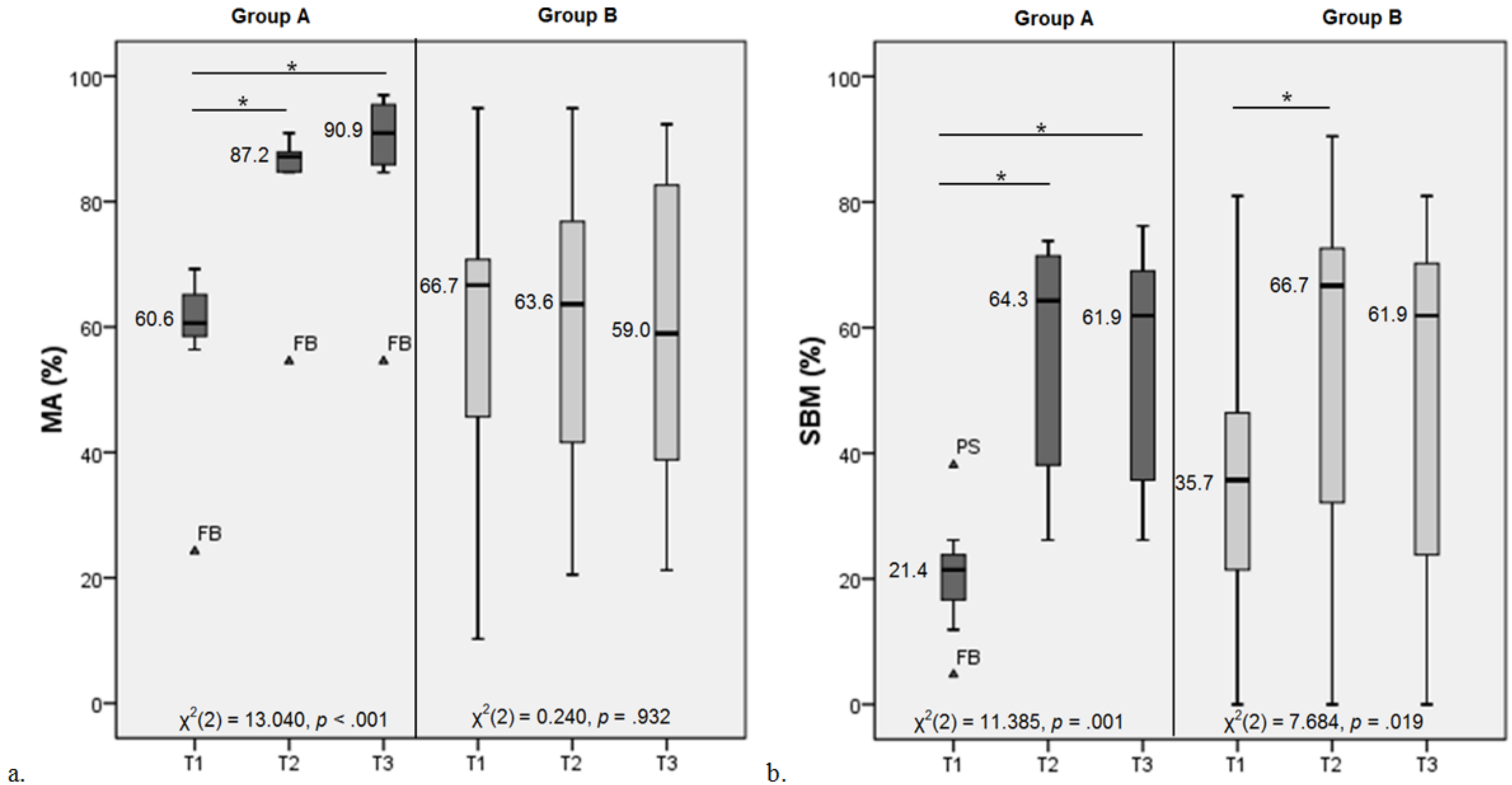


Figure 8.2 The subscores of the Water Orientation Test Alyn 2 at T_1, T_2, T_3 and the results of the statistical analyses of the changes over time (T_1, T_2, T_3). Boxplots for the results of Group A (*n* = 7), participating in the swimming intervention between T_1 and T_2, and of Group B (*n* = 7), the control group, are displayed; a. Mental adjustment (MA) subscale; b. Skills, balance control and movement (SBM) subscale. Note that Participants FB and PS have outlying scores. * *p* < .05.

The analysis of the data from the BA-design of Group B (n = 7), participating in the swimming intervention following the control period (Figure 3.4b), agreed with the main analysis that the improvement in swimming skills over a 10-week swimming intervention was larger than over the control period. The total score and the MA subscore changed significantly more over the intervention period (T_3–T_4) than over the control period (T_1–T_2). The total score increased by a median of 29.6% over the intervention period, compared to 6.2% over the control period (Z = -2.028, p = .047). The MA subscore increased 24.2% over the intervention period compared to a median change of 0.0% over the control period (Z = -2.028, p = .047). The differences for both the MA subscore and the total score represent large ES, r = 0.54. Changes in the SBM subscore were not significantly different between the swimming (21.4% absolute increase) and the control period (16.7% absolute increase), Z = -1.527, p = .141; however, the difference represents a medium ES, r = 0.41.

Participants in Group A (n = 7) retained their swimming skills after completing the swimming intervention. After the 15- and 20-week follow-up period that followed the completion of the swimming intervention, the total score and both subscores of the WOTA 2 remained significantly different from the baseline values. The differences between baseline and follow-up represented large ES, as shown in Table 8.3. For the total score, an increase in swimming skills was retained ranging from a 11.1% to a 42.7% absolute increase after 15 weeks of follow-up, and with an increase ranging from a 12.4% to a 45.3% absolute increase at 20 weeks of follow-up.

Table 8.3 Results of the statistical analyses of the Water Orientation Test Alyn 2 (%) comparing baseline scores and the scores after a 15- and 20-week follow-up period in Group A (*n* = 7).

Subscales	15-week follow-up period				20-week follow-up period			
	Median difference (IQR)	*Z*	*p*	*r*	Median difference (IQR)	*Z*	*p*	*r*
TOT	39.5 (20.7)	-2.371*	.016	0.63	33.3 (24.0)	-2.366*	.016	0.63
MA	33.3 (7.6)	-2.384*	.016	0.63	28.2 (15.2)	-2.371*	.016	0.63
SBM	45.2 (29.8)	-2.371*	.016	0.63	38.1 (31.0)	-2.366*	.016	0.63

Note. Absolute median change scores from baseline to 15 weeks follow-up and to 20 weeks follow-up, and the results of the Wilcoxon signed-rank test are displayed.
TOT = total score; MA = mental adjustment subscore; SBM = skills, balance control and movement subscore. IQR: inter quartile range. *r* represents the effect size estimate for the Wilcoxon signed-rank test.* $p < .05$.

8.1.1.1 Clinical significance

The minimal detectable change based on the confidence interval of 95% for the WOTA 2 is 11.5 points, or 14.2%, according to Tirosh et al. (2008). In the present study all youth but two improved by more than the minimal detectable change (14.2%) over the swimming period. One participant of Group A improved by 12.4% and one participant in Group B, with a baseline score of 87.7%, improved by 8.6% over the swimming period. After 15 and 20 weeks of follow-up 6 of the 7 participants of Group A retained a change score higher than the minimal detectable change.

8.1.2 Influence of the confounding variables

During the intervention programme, one adolescent of Group A swam at school once every 21 days, but the WOTA 2 scores did not increase more than for the other participants of Group A, as the scores of the adolescent increased by the median score of 33.3%. Three participants of Group B swam at school every fortnight throughout the study period. The WOTA 2 total scores of 2 of these participants increased by a substantial amount (17.3% and 24.0%) over the control period (T_1–T_2). The WOTA 2 total score of the third participant increased by less than the median for this group. Nevertheless, 2 of the 3 participants, who performed extra swimming at school, increased more over the swimming period (T_3–T_4) than over the control period; and one participant, that had a 24.0% absolute increase over the control period, improved by a further 16.0% over the swimming programme. During the period following completion of the swimming intervention in Group A, 3 participants swam at school less than, or equal to, once every 14 days and one participant went swimming with his father regularly. These 4 participants improved (n = 1), deteriorated (n = 2) or did not change (n = 1) over this follow-up period.

The WOTA 2 total score of the child from Group A that was given a botulinum toxin A injection between T_3 and T_4 deteriorated by 9.3%. This was the largest decrease for the participants of Group A over this period. However, the improvement since baseline (T_1) was still significant (20.0%). The participant that underwent brain surgery between T_3 and T_4 did not deteriorate over this period.

8.2 Discussion

The purpose of the present study was to investigate the effect of a 10-week swimming programme on mental adjustment in the water and swimming skills in ambulatory youth with CP, and to investigate the retention after a 5- to 20- week follow-up period. The findings show that swimming skills in children and adolescents with CP (Gross Motor Function Classification System levels I to III) improved significantly more over the 10-week swimming intervention than over the control period and that these improvements were retained after 20 weeks of follow-up. Since motor proficiency is associated with physical activity, fitness and participation levels (Burns et al., 2009; Kang et al., 2010; Wrotniak et al., 2006), this has important implications for the vicious cycle of deconditioning.

Both the mental adjustment subscore (MA) and the skills, balance control and movement subscore (SBM) increased significantly more in Group A, participating in the swimming intervention (T_1–T_2) than in Group B, the control group. The improvement in swimming skills after the swimming intervention supports previous research (Dimitrijević et al., 2012; Getz et al., 2007; Hutzler, Chacham, Bergman, & Reches, 1998; Hutzler, Chacham, Bergman, & Szeinberg, 1998; Jorgić, Dimitrijević, Aleksandrović, et al., 2012). However, in these studies no control group had performed the aquatic tests. In the present study both the MA score and the SBM score had increased after swimming. The SBM score in Group A increased (36%) more than the MA score (24%) for this group. These results are comparable to previously published results. The RCT of Dimitrijević et al. (2012) reported a mean increase of 31% in the WOTA 2 total score, with an increase of 29% for the MA score and an increase of 33% for the SBM score after swimming 45 min, twice a week, for 6 weeks. One case series design (Jorgić, Dimitrijević, Aleksandrović, et al., 2012) reported an increase of 25% for the total score, with the MA score improving by 27% and the SBM score by 34% after swimming 45 min, twice a week, for 6 weeks. The larger increase for the SBM score than for the MA score might be explained by the lower baseline values for this subscale.

The present study included a control group that performed the aquatic test, in contrast to the previous studies reported in the extant literature. Over this 10-week control period of Group B (T_1–T_2) there was a median increase of 6% in the total score of the WOTA 2, mainly due to a significant increase in the SBM part of the scale (17% increase). Closer inspection of the data for individuals revealed that the SBM score improved by a 10% to a 36% absolute increase over the 10-week control period for 5 participants of Group B, and remained unchanged for the 2 other participants. These large improvements are mainly due to two 7 to 8 year-old participants improving by more than 30% for the SBM score over the control period (T_1–T_2). An explanation for this improvement might be associated with the learning effect during the assessment at baseline, and with the participation of the 2 children in swimming classes provided during their school programme every 14 days. Nevertheless, there was a significantly larger improvement in Group A over the swimming intervention, than the improvement in Group B over the control period. This implies that greater levels of swimming, such as in the intervention programme offered in the present study, are worthwhile.

Group B, participating in the intervention programme following the control period (T_3–T_4), improved significantly more over the swimming programme than over the control period for the total score and the MA score. Although not reaching statistical significance, the SBM score increased more over the swimming intervention than over the control period of Group B and represented a medium ES. The absence of reaching significance might be due to the high baseline levels for the SBM subscale obtained after the control period. All participants of Group B, except for the two 7 to 8 year-old children mentioned in the previous paragraph, increased more for the SBM subscale over the swimming period than over the control period. The increase for the MA subscore was larger than the increase for the SBM subscore after Group B's swimming intervention, which is in contrast with the results of Group A and previously published studies (Dimitrijević et al., 2012; Jorgić, Dimitrijević, Aleksandrović, et al., 2012). This inconsistent result might be explained by the large baseline values for the SBM score obtained after the control period, and by a large individual increase (59%) for the MA subscale of the adolescent with very low baseline levels for the MA subscale (10%) due to fear or anxiety towards the water. These results imply that the scores on both subscales of the WOTA 2 increased due to swimming. Whether mental adjustment to the water or motor skills in the water increased the most, was depended on the baseline level, as well as on the presence of fear or anxiety towards the water.

For both subscales the improvements were retained with significance for 15 and 20 weeks after completing the swimming programme. No comparisons could be made with other studies due to the lack of follow-up periods longer than 3 weeks. Dimitrijević et al. (2012) reported no decrease for the WOTA 2 total score between the post-test and 3 weeks of follow-up. The strong retention existing in the present study implies that the swimming skills were learned and consolidated during the 10-week intervention. Additionally, this retention can facilitate continued engagement in aquatic activities.

The present study confirms that swimming skills improve significantly over a 10-week swimming programme in ambulatory youth with CP, and shows that the skills improve significantly more than over the control period. Swimming skills improved by more than the minimal detectable change in 12 of the 14 participants. The findings of the present study could encourage youth with CP to learn how to swim, as one of the perceived barriers reported by youth with CP to engaging in physical activity is the belief that learning a motor skill is too time consuming (Verschuren et al., 2012). Moreover, the retention of swimming skills enables the participants to engage in other physical activities performed in the water, and to engage in a greater variety of activities with friends (Kang et al., 2010).

Limitations of the study and suggestions for future research are discussed in sections 11.3 and 11.4 of the 'General Discussion' chapter.

Chapter 9: Results and discussion of Research Question 5 – Level of enjoyment

9.1 Results

All participants (*N* = 14) rated how much they had enjoyed each swimming session on a scale of 1 to 5.

As the data of Table 9.1 show, the median level of enjoyment score was 5 (IQR = 0) for each group separately. All individuals but one had a median score of 5 (i.e. the maximum score), indicating that the swimming sessions were enjoyed ‘very much’. One child from Group A reported a median score of 3, indicating that the sessions were enjoyed ‘a little bit’.

Table 9.1 Levels of enjoyment of the swimming intervention.

Initials	Group	Enjoyment (1 – 5)
JVB		5 (0.0)
FB		3 (2.0)
FT		5 (0.0)
PS	A	5 (0.0)
EG		5 (1.0)
RR		5 (0.3)
FS		5 (1.0)
Median		5 (0.0)
AB		5 (0.0)
FP		5 (0.3)
LP		5 (0.0)
KR	B	5 (0.0)
SD		5 (1.0)
JS		5 (1.0)
SV		5 (0.0)
Median		5 (0.0)

Note. Values are medians (inter quartile ranges) of the completed sessions for each participant, and group medians.

9.2 Discussion

The aim of the present study was to assess the level of enjoyment of the swimming sessions provided to youth with CP, as one of the perceived barriers reported by youth with CP to engaging in physical activity is the perception of physical activity and sports as not being fun. The present study was the first to report the level of enjoyment of the exercise intervention experienced by youth with CP. All participants but one reported enjoying the swimming sessions 'very much' (maximum score of 5). By participation in the swimming programme the children and adolescents experienced that sport and exercise programmes, in this case swimming, can be fun. Therefore, a swimming programme such as the one offered in this study has the ability to eliminate one of the barriers to engaging in physical activity (Verschuren et al., 2012).

Since enjoyment and motivation are important facilitators for engaging in physical activity, adhering to therapy, and sustaining a physically active lifestyle (Buffart et al., 2009; Redmond & Parrish, 2008; Riner & Sellhorst, 2013), the findings of the present study are of importance. The suggested positive relationship between enjoyment and adherence is supported by the combination of high enjoyment and high attendance rates found in the present study. Attendance to the swimming programme (section 4.3) was 95% for the entire sample ($N = 14$). Only one other study investigating an aquatic programme in youth with CP reported an attendance rate, which was 80% (Fragala-Pinkham et al., 2008). Other exercise programmes reported attendance rates of 90% for a stationary cycling programme (Demuth et al., 2012), 93% for an aerobic and anaerobic exercise programme (Verschuren et al., 2007) and 78% and 72% for an over-ground walking practice and a partial body weight treadmill training programme (Willoughby et al., 2010). The systematic reviews on the effects of exercise programmes in youth with CP (Butler, Scianni, & Ada, 2010; Rogers, Furler, Brinks, & Darrah, 2008; Verschuren et al., 2008) did not report the attendance rates of the programmes. Furthermore, only one child withdrew from the study, before the second measurement occasion. The child that withdrew was assigned to the control group, and therefore was not enrolled in the swimming programme yet. More importantly, none of the participants dropped out during the intervention. Other studies investigating aquatic programmes in youth with CP reported dropout rates varying from no dropout (Chrysagis et al., 2009) to a dropout of 2 or more participants (Ballaz et al., 2011; Dimitrijević et al., 2012; Fragala-Pinkham et al., 2008; Fragala-Pinkham et al., 2010; Getz et al., 2012), with the highest dropout of 17 out of 40 participants in the randomised controlled trial of Őzer et al. (2007). Other exercise programmes reported a dropout of 3 of the 68 participants during the baseline measurement (Verschuren et al., 2007), 8 dropouts between baseline and post-test of the 33 participants (Willoughby et al., 2010) and 2 dropouts of the 33 participants in the experimental group between baseline and post-test (Demuth et al., 2012). The low dropout rate in the present study implies that the motivation to complete the swimming programme was high.

Since adherence to the swimming programme was high, the reported levels of enjoyment of the swimming programme were high and none of the participants withdrew during the intervention, we can conclude that the intervention was experienced as fun, and motivated the youth to continue participating. Additionally, motivation to continue engaging in physical activity was present since all but one participants engaged in swimming after the completion of the programme (results are presented in Chapter 10). This finding is in agreement with the literature, reporting that motivation and fun are facilitators for engagement in physical activity, and on-going sport commitment (Martin, 2006).

These findings, however, cannot be extrapolated to all youth with CP. The children and adolescents were recruited based on a convenience sample, therefore it was expected that all of the participants would have been keen on swimming. One adolescent girl had experienced swimming negatively and was therefore not keen on taking part in the swimming intervention. After an introductory session without obligations, she independently decided to participate in the study. This case study shows the value of introductory sessions without obligations or cost to introduce the activity and to allow the potential participant to perceive the possible enjoyment and to experience the approach of the (swimming) instructor.

In conclusion, swimming is a physical activity that was highly enjoyed by the children and adolescents with CP participating in the present study. This enjoyment can help eliminating the barriers to engaging and to adhering to physical activity. The results from the present study suggest that youth with CP would benefit from the opportunity to engage in a swimming programme to enable them to experience the high levels of enjoyment when exercising. Swimming is therefore a recommended physical activity for children and adolescents with CP (with the ability to walk) to combat the vicious cycle of deconditioning, and should be promoted. Highly recommended is to assess the level of enjoyment in any intervention programme, since it closely relates to adherence. Physical activity and sport programmes should be promoted to youth with CP with trial and introductory sessions provided in community-based settings in collaboration with physical therapists, as a lack of information sustains the perception that physical activity and sport programmes are not fun.

Chapter 10: Results and discussion of Research Question 6 – Participation in recreational, active-physical, social and skill-based activities

10.1 Results

The participants completed the Children's Assessment of Participation and Enjoyment (CAPE) at baseline and one year later. The scale assesses five dimensions of participation (diversity, intensity, 'with whom', 'where', and the enjoyment) for 55 activities. The scale questions the participation in these activities over the previous 4 months. The pre-test questionnaires were completed between October and December 2011 and the post-test questionnaires were completed between October and November 2012.

The cohort had participated in 25 of the 55 activities in the 4 months prior to baseline, as the total score for diversity in Table 10.1 shows. The activities that the children and adolescents performed the most, at baseline, are displayed in Table 10.2. The highest intensities (frequency of participation, calculated as the intensity for each activity divided by the number of possible activities) for these tasks were reported for passive activities such as 'watching TV', 'listening to music' and 'playing computer or video games'. None of the youth participated in the following activities: 'hanging out', 'martial arts', 'swimming class', 'gymnastics', 'team sports', 'snow sports', 'volunteer work', 'a paid job'.

The diversity of activities in which the cohort had participated in the 4 months prior to baseline, was higher in the informal domain (spontaneous, involving little planning, initiated by the child) than in the formal domain (structured, involving rules or goals). Participation diversity in recreational and social activities was higher than in active-physical and skill-based activities (Table 10.3). At baseline, the youth had participated in all 55 activities with a mean intensity (the frequency of participation) of 1.95 out of 7, which represents once to twice in the previous 4 months (Table 10.1). Differences for participation intensity between domains and activity types were similar to the differences described for participation diversity and are presented in Table 10.4. The results for the total score, at baseline and for the changes over one year, are presented in Table 10.1. The results for the informal and formal domain, and the five activity types, are shown in Tables 10.3 – 10.7, presented later in this section.

Table 10.1 Baseline values and the changes over one year for the total scores of the participation dimensions of the Children's Assessment of Participation and Enjoyment.

Total score (*N* = 14)	Baseline Mean (*SD*)	Change over one year Mean (*SD*)	95% Confidence Interval of the change	*t*(13)	*p*	*d*
Diversity (0 – 55)	24.93 (4.63)	.071 (5.11)	-2.88 – 3.02	0.052	.959	0.01
% Diversity	45.32 (8.42)	.131 (9.28)	-5.23 – 5.49	0.053	.959	
Intensity (1 – 7)	1.95 (0.44)	.030 (0.37)	-0.19 – 0.25	0.300	.769	0.18
With whom (1 – 5)	2.39 (0.40)	.212 (0.42)	-0.03 – 0.46	1.871	.084	0.48
Where (1 – 6)	2.72 (0.43)	.155 (0.59)	-0.19 – 0.50	0.978	.346	0.28
Enjoyment (1 – 5)	4.49 (0.42)	-.128 (0.34)	-0.33 – 0.07	-1.396	.186	0.31

Note. The % Diversity is the number of activities participated in divided by the number of possible activities to participate in (55), multiplied by 100. The change over one year was calculated as the post-test value minus the pre-test value. *d* represents the effect size for the change over one year.

To the question 'With whom do you do this activity most often?' a mean sample score of 2.39 out of 5 was recorded (Table 10.1). This number indicates carrying out activities with family. For active-physical and skill-based activities and activities of the formal domain higher scores were reported than in the other activity types, indicating that these activities were performed with friends and others rather than with family or alone (Table 10.5). Children and adolescents mostly participated in activities at home or in their neighbourhood. For the activities of the formal domain, active-physical and skill-based activities higher scores were reported than for the other activities, indicating that these activities were more community-based than home-based (Table 10.6). The youth enjoyed participating in the activities 'very much' (4.49 out 5). High levels of enjoyment were experienced during participation in all domains and activity types, with the lowest enjoyment reported for self-improvement activities. Youth enjoyed participation in skill-based activities the most (Table 10.7).

Table 10.2 The top 20 of activities participated in at baseline.

	Activity	*N* = 14	% *N*	Intensity (1 – 7)
1	Watching TV or a rented film	14	100.0	6.9
2	Visiting	14	100.0	3.3
3	Listening to music	13	92.9	6.8
4	Shopping	13	92.9	4.2
5	Computer/video games	13	92.9	6.5
6	Going on a full-day outing	12	85.7	2.4
7	Homework	12	85.7	5.8
8	Playing board or card games	12	85.7	3.3
9	Doing pretend or imaginary play	12	85.7	5.2
10	Reading	11	78.6	6.3
11	Crafts, drawing, colouring-in	11	78.6	5.5
12	Going for walk/hike	11	78.6	4.2
13	Talking on the phone	11	78.6	5.0
14	Going to a party	11	78.6	2.4
15	Cycling, in-line skating, or skateboarding	11	78.6	4.8
16	Playing with toys/things	10	71.4	6.3
17	Playing with pets	10	71.4	6.1
18	Entertaining others	10	71.4	2.8
19	Doing a chore	9	64.3	5.4
20	Making food	8	57.1	3.3
	Going to a live event	8	57.1	1.6
	Going to the cinema	8	57.1	1.5
	Dancing	8	57.1	4.4

Note. Place number 20 is shared by 4 activities.

One year after baseline, none of the total scores differed significantly from baseline (Table 10.1). However, all total scores had increased, except for the level of enjoyment. 'With whom' youth participated improved by a medium effect size (ES) (Table 10.1). The following sections present the changes over one year for each dimension of participation, specified for the various domains and activity types.

10.1.1 Change in participation diversity

Although significance was not reached, there was a small increase in participation diversity for the formal domain (absolute increase of 1.4%), active-physical activities (5.5%) and skill-based activities (2.9%) (Table 10.3). The number of activities of the informal domain and the other activity types, in which they participated, decreased non-significantly. The difference between pre-test and post-test for participation diversity of the active-physical activities represents a small to medium ES.

Table 10.3 The percentage of diversity at baseline and the change over one year for the two domains and five activity types of the Children's Assessment of Participation and Enjoyment.

% Diversity (N = 14)	Baseline Mean (SD)	Change over one year Mean (SD)	95% Confidence Interval of the change	t(13)	p	d
Informal	56.07 (9.7)	-0.36 (11.0)	-6.73 – 6.02	-0.121	.906	0.03
Formal	16.67 (8.2)	1.43 (10.8)	-4.83 – 7.69	0.493	.630	0.13
Recreational	69.05 (21.3)	-1.19 (17.6)	-11.33 – 8.95	-0.254	.804	0.06
Active-physical	19.23 (8.4)	5.50 (14.6)	-2.94 – 13.93	1.408	.183	0.47
Social	67.86 (16.3)	-2.14 (18.1)	-12.57 – 8.28	-0.444	.664	0.12
Skill-based	25.71 (13.4)	2.86 (14.4)	-5.44 – 11.16	0.744	.470	0.19
Self-improvement	47.86 (14.2)	-5.71 (18.7)	-16.51 – 5.08	-1.144	.273	0.31

Note. Changes over one year were calculated as the post-test value minus the pre-test value. d represents the effect size for the change over one year.

10.1.2 Change in participation intensity

Participation intensity (frequency of participation, calculated as the intensity for each activity divided by the number of possible activities) increased non-significantly for the formal domain (increase of 0.114 or 1.6% relative increase to baseline), active-physical activities (3.8%), social activities (0.3%), and skill-based activities (2.8%), and decreased non-significantly for the informal domain, the recreational and the self-improvement activities (Table 10.4). The difference between pre-test and post-test for participation intensity in the active-physical activities represents a medium ES. However, all changes in participation intensity were smaller than the smallest detectable changes reported by Bult et al. (2010) that range from 1.14 to 1.86 for the five activity types.

Table 10.4 Baseline values and change over time for the participation intensity of the two domains and five activity types of the Children's Assessment of Participation and Enjoyment.

Intensity (1 – 7)	Baseline Mean (*SD*)	Change over one year Mean (*SD*)	95% Confidence Interval of the change	*t*(13)	*p*	*d*
Informal	2.45 (0.54)	-.002 (0.44)	-0.26 – 0.25	-0.015	.988	0.00
Formal	0.61 (0.40)	.114 (0.49)	-0.17 – 0.40	0.877	.396	0.25
Recreational	3.57 (1.26)	-.107 (0.75)	-0.54 – 0.33	-0.534	.602	0.09
Active-physical	0.72 (0.33)	.264 (0.55)	-0.06 – 0.58	1.788	.097	0.54
Social	2.31 (0.53)	.021 (0.74)	-0.41 – 0.45	0.108	.916	0.03
Skill-based	1.06 (0.59)	.193 (0.65)	-0.18 – 0.57	1.106	.289	0.33
Self-improvement	2.12 (0.55)	-.264 (0.53)	-0.57 – 0.04	-1.876	.083	0.36

Note. Changes over one year were calculated as the post-test value minus the pre-test value. *d* represents the effect size for the change over one year.

10.1.3 Change in 'with whom' youth participate

With whom the children and adolescents participated in activities of the formal domain had changed significantly one year after baseline. The score increased by 1.05 points out of 5, representing a large ES, and indicating participation in formal activities with friends and others rather than with family. The participants performed the activities of the informal domain and all the activity types slightly less with family or alone than at baseline (Table 10.5). The difference between pre-test and post-test for the skill-based activities represents a medium ES.

Table 10.5 Baseline values and changes over time for the two domains and five activity types of the Children's Assessment of Participation and Enjoyment for the dimension 'with whom'.

With whom (1 – 5)	Baseline Mean (*SD*)	Change over one year Mean (*SD*)	95% Confidence Interval of the change	*t*(13)	*p*	*d*
Informal	2.26 (0.42)	0.13 (0.47)	-0.15 – 0.40	0.989	.341	0.26
Formal *t(12)*	3.50 (0.88)	1.05 (0.96)	0.46 – 1.63	3.914**	.002	1.42
Recreational	2.06 (0.58)	0.09 (0.52)	-0.21 – 0.39	0.633	.538	0.14
Active-physical	2.76 (0.93)	0.03 (1.28)	-0.71 – 0.77	0.080	.938	0.03
Social	2.47 (0.54)	0.14 (0.49)	-0.14 – 0.43	1.075	.302	0.25
Skill-based *t(12)*	3.48 (1.14)	0.51 (1.08)	-0.14 – 1.16	1.706	.114	0.54
Self-improvement	2.12 (0.57)	0.03 (0.99)	-0.54 – 0.60	0.125	.902	0.05

Note. Changes over one year were calculated as the post-test value minus the pre-test value. *d* represents the effect size for the change over one year. ** $p < .01$.

10.1.4 Change in 'where' youth participate

The place where children and adolescents participated in the activities did not change significantly one year after baseline. For the formal domain and the self-improvement activities a small change towards more home-based participation was reported, while all others showed a slight change towards more community-based participation (Table 10.6). Differences between pre-test and post-test were the greatest in the active-physical and skill-based activities and represent small to medium ES.

Table 10.6 Baseline values and change over time of the two domains and various activity types of the Children's Assessment of Participation and Enjoyment for the dimension 'where'.

Where (1 – 6)	Baseline Mean (*SD*)	Change over one year Mean (*SD*)	95% Confidence Interval of the change	*t*(13)	*p*	*d*
Informal	2.52 (0.47)	.190 (0.64)	-0.18 – 0.56	1.108	.288	0.32
Formal *t(12)*	4.35 (1.39)	-.129 (0.75)	-0.58 – 0.32	-0.623	.545	0.11
Recreational	2.01 (0.77)	.101 (0.90)	-0.42 – 0.62	0.420	.681	0.13
Active-physical	3.59 (1.10)	.535 (1.63)	-0.41 – 1.48	1.225	.242	0.45
Social	3.03 (0.43)	.041 (0.86)	-0.46 – 0.54	0.177	.863	0.06
Skill-based *t(12)*	3.53 (1.29)	.230 (0.85)	-0.29 – 0.75	0.968	.352	0.38
Self-improvement	2.47 (0.56)	-.046 (0.83)	-0.52 – 0.43	-0.208	.838	0.06

Note. Changes over one year were calculated as the post-test value minus the pre-test value. *d* represents the effect size for the change over one year.

10.1.5 Change in participation enjoyment

How much the children and adolescents enjoyed the activities in which they participated, did not differ significantly between baseline and one year after baseline. The enjoyment reported for all domains and activity types decreased slightly; however, youth still enjoyed participation in all activities ‘much’ to ‘very much’, except for the participation in self-improvement activities, which was enjoyed ‘a little’ to ‘much’ (Table 10.7).

Table 10.7 Baseline values and changes over time in participation enjoyment of the two domains and various activity types of the Children’s Assessment of Participation and Enjoyment.

Enjoyment (1 – 5)	Baseline Mean (*SD*)	Change over one year Mean (*SD*)	95% Confidence Interval of the change	*t*(13)	*p*
Total	4.49 (0.42)	-.128 (0.34)	-0.33 – 0.07	-1.666	.186
Informal	4.48 (0.42)	-.166 (0.37)	-0.38 – 0.05	-1.144	.120
Formal *t(11)*	4.53 (0.58)	-.235 (0.71)	-0.69 – 0.22	-0.514	.277
Recreational	4.55 (0.59)	-.093 (0.68)	-0.48 – 0.30	-1.055	.616
Active-physical	4.61 (0.61)	-.232 (0.82)	-0.71 – 0.24	-0.879	.311
Social	4.60 (0.39)	-.078 (0.33)	-0.27 – 0.11	-1.798	.396
Skill-based *t(12)*	4.71 (0.43)	-.290 (0.58)	-0.64 – 0.06	-1.565	.097
Self-improvement	3.97 (0.91)	-.257 (0.61)	-0.61 – 0.10	-1.666	.142

Note. Changes over one year were calculated as the post-test value minus the pre-test value.

10.1.6 Participation in aquatic activities

In the 4 months preceding baseline none of the youth participated in a formal swimming class, and only 7 youth participated in informal aquatic activities, with a maximum intensity of once per week. In the 4 months preceding the post-test, one year later, 4 youth participated in a formal swimming programme, up to once per week, and 13 youth participated in informal aquatic activities (up to 2 – 3 times per week). One adolescent girl did not participate in informal aquatic activities due to restrictions following orthopaedic surgery.

10.1.7 Influence of the confounding variables

The participant that underwent orthopaedic surgery and consequently was not allowed to undertake several activities during the 4-month period over which the CAPE assesses the participation dimensions, did not have lower change scores than the sample mean, except for a small decrease in enjoyment and 'where' the activities took place. The participant that changed schools reported a large increase in the diversity and intensity of participation, performed activities more with friends and others than with family or alone, and performed activities more in the broader community than at home. When the latter participant was excluded from the analyses, changes for 'where' and 'with whom' the sample performed activities remained at a similar level, and the changes for participation diversity and intensity of the active-physical activities were slightly smaller.

10.2 Discussion

The purpose of the present study was to investigate the change in participation in recreational, active-physical, social and skill-based activities over one year, during which all youth participating in the study received a 10-week swimming programme.

Participation diversity and intensity for the active-physical activities improved slightly and the changes represented medium ES. Since these activities are the most community-based, physically active as well as performed with friends rather than with family, these are the activities to target when addressing the vicious cycle of deconditioning. Additionally, youth participated significantly more with friends and others in activities of the formal domain than at baseline. Although not reaching significance, one year after baseline, youth performed skill-based activities more with friends and others than with family or alone (medium ES), and performed active-physical and skill-based activities slightly more in community-based places than at home (small to medium ES). Since youth with CP are known to participate in activities with family at home rather than with friends in the community (Engel-Yeger et al., 2009), increases in participation with friends in the community are of importance for the child's development. Additionally, the participants engaged in more swimming and aquatic activities than at baseline.

Participation diversity increased for the formal domain (1.4%), active-physical activities (5.5%) and skill-based activities (2.9%) and participation intensity increased for active-physical activities (3.8%) and skill-based activities (2.8%); all changes over one year represented small to medium ES. The difference between pre-test and post-test for participation intensity in the active-physical activities represents a medium ES. However, all changes in intensity were smaller than the smallest detectable changes reported by Bult et al. (2010). No control data were obtained in the present study, so it is not possible to compare to the change over one year for youth with CP that did not participate in a swimming programme. Nevertheless, it is known that participation diversity and intensity in leisure activities decreases as youth grow older (Law et al., 2006; Shikako-Thomas et al., 2013). This decrease was also reported in the intervention study conducted by Verschuren et al. (2007) who found that participation intensity in the control group decreased over a period of 8 months. The small improvements for participation diversity and intensity in the present study suggest that taking part in the swimming programme may have prevented deterioration in participation.

For the present sample participation diversity and intensity at baseline were lower in activities of the formal domain than in activities of the informal domain, and were lower in the active-physical and skill-based activities than in the recreational and social activities. Lower participation diversity in active-physical and skill-based activities than in other activity types, is in agreement with the findings of Imms et al. (2008). The top 5 of activities that all children and adolescents participated in were all, except for shopping, passive leisure activities; similar to the results of Imms et al. (2008). Some of the activities (watching TV, listening to music, playing computer games) were carried out almost once or more per day. This finding is not different from the typical developing youth (Imms et al., 2008), but does confirm the problem of physical inactivity. The baseline values for participation intensity of the present study were slightly lower than those of the Dutch 6 to 18 year-old typically developing youth of the sample of Bult et al. (2010) for all activity types, except for recreational activities. In particular, considerably lower participation intensity values were present for the active-physical activities compared to the sample of Bult et al.

(2010). This confirms the need for facilitating participation in active-physical interventions.

Participation in formal, active-physical and skill-based activities at pre-test and at post-test occurred more with friends and others than with family or alone compared to the other activity types, and participation in these activities was more community-based than in other activity types. This finding is in agreement with the results of Shikako-Thomas et al. (2013), investigating participation levels of adolescents with CP. Additionally, youth experienced high levels of enjoyment when participating in formal, active-physical and skill-based activities.

One year after baseline, the children and adolescents of the present sample participated significantly more with friends and others than with family or alone in formal activities. In skill-based activities a similar improvement was found, representing a medium ES. The place and enjoyment of participation did not change significantly over one year. However, skill-based activities and active-physical activities were slightly more community-based (small to medium ES). The changes over time for these participation dimensions have not been reported in previous studies in the extant literature and therefore, no comparison could be made.

The significant change over time for participation in formal activities with friends rather than with family or alone is important for the social development of the child. This finding implies that learning the skill of swimming introduces youth to a formal activity with a more extended circle of people, and confirms the research of Kang et al. (2010), which indicated that youth who scored higher on sports and physical function participated in more activities with friends.

In conclusion, at baseline, participation diversity and intensity were lower in the formal than in the informal domain, and lower in the active-physical and skill-based activities than in the recreational and social activities. At baseline, participation in these formal, active-physical and skill-based activities occurred more with friends and others than with family or alone, and was more community-based than in the other activity types. Hence, at the start of this study, the participating youth were particularly restricted in their participation in formal, active-physical and skill-based activities at the community with peers. One year after baseline that included the swimming programme, the children and adolescents with CP performed the activities of the formal domain and skill-based activities more with friends than at baseline. Additionally, small changes in diversity and intensity of participation in active-physical activities occurred. These results are promising in view of the deterioration in participation with increasing age reported in the literature for youth with CP. The findings suggest that swimming encouraged participation in activities of the formal domain, active-physical and skill-based activities, and facilitated youth to engage in aquatic activities. A next step would be to investigate the effect of swimming on participation including a control group.

Limitations of the study and suggestions for future research are discussed in sections 11.3 and 11.4 of the 'General Discussion' chapter.

Chapter 11: General discussion

The aim of this thesis was to investigate the effect of swimming on a complex set of interrelated factors that are associated with the physical inactivity and low fitness levels that are present in youth with cerebral palsy (CP), as was displayed in Figure 1.3 of Chapter 1. These factors are pain intensity, perceptions of fatigue, walking ability, coordination, functional independence, self-perception, quality of life (QoL) and participation in leisure activities. There is a paucity of studies in the literature investigating the influence of swimming on the various impairments, activity limitations and participation restrictions encountered by youth with CP. The present study was the first randomised controlled trial to investigate the effect of swimming in 7 to 17 year-old youth with CP with the ability to walk (with or without hand-held mobility devices), on pain, fatigue, walking ability, bilateral and upper limb coordination, perceived competence and global self-worth, and QoL. Whether participation in a 10-week swimming programme influenced participation levels in leisure activities over one year was investigated using a pre-test – post-test design. The latter was the first study including more than two participants with CP that investigated the effect of swimming on participation. Additionally, the effects of participating in a 10-week swimming intervention on mental adjustment in the water and swimming skills, as well as the retention over a 20-week follow-up period were assessed, as one of the barriers to engaging in physical activity is the perception that learning a motor skill is too time-consuming (Verschuren et al., 2012). Moreover, the enjoyment of the swimming intervention was assessed, as fun and enjoyment are facilitators for engaging in physical activity, for improving adherence to physical activity and for continuing committing to sport (Buffart et al., 2009; Martin, 2006; Redmond & Parrish, 2008). Adherence to physical activity is essential to sustain a healthy lifestyle.

11.1 Overview of findings

The present study showed that walking distance at maximum walking speed improved significantly more over the swimming intervention, than over the control period, while pain intensity and perceptions of fatigue were not affected adversely. Medium to large effect sizes were found for the differences between the changes in walking distance over the swimming intervention (in both Group A and Group B) and changes over the control period (Group B). The increase in walking distance was retained after 20 weeks of follow-up that followed the completion of the swimming programme in Group A. After this follow-up period, the scores of Group A differed significantly from baseline, with 5 of the 7 participants of this group increasing by more than 13 m, which was deemed clinically relevant (McDowell et al., 2009). Since motor functioning can limit the ability of youth to keep up with peers in the community (Kang et al., 2010; Palisano et al., 2009), the improvement in walking speed is important for participation in physical and daily activities with friends. In the present study, feelings of fatigue occurred rarely in the month prior to baseline or prior to the post-intervention test. Over the 15-week control period ($T_1 – T_3$) fatigue increased significantly in Group B and differences between the changes over the control period and changes over the swimming period represented medium effect sizes. It is pertinent that engaging in the swimming programme did not increase levels of pain or fatigue and that a slight reduction of feelings of fatigue was reported by the participants, since this eliminates the perception of pain and fatigue to increase due to exercise (Verschuren et al., 2012).

Upper limb coordination and functional independence in mobility and social functioning might improve more over a swimming intervention than over a control period. Statistical significance was reached for functional independence in social functioning and the differences between the changes over the 10-week swimming period and changes over the control period for upper limb coordination and functional independence in mobility represented medium effect sizes. For upper limb coordination 5 of the 7 participants of Group A improved by more than the minimal important difference over the swimming intervention. The present study supported the previous findings reporting improvements in social functioning and mobility after an aquatic intervention (section 6.2). Additionally, the improvement in social functioning was maintained with large and medium effect sizes after 15 and 20 weeks of follow-up. The improvement of functional independence in social functioning is important with respect to the engagement in the community. The improvements in upper limb coordination and functional independence in mobility are important with regard to the influence of motor proficiency on physical activity, fitness and participation (Bult et al., 2013; Burns et al., 2009; Kang et al., 2010; Okely, 1999; Wrotniak et al., 2006). The improvements in social functioning and mobility were expected, since the swimming programme involved participating in activities with peers, but also with instructors, and other persons present in the swimming pool. Similarly, independence in mobility was expected to improve, since walking ability had improved and an extra effort was made to come to the swimming pool, enter the pool and walk around in the pool. Also functional independence in self-care was expected to improve, as the programme was offered at a community swimming pool and youth were encouraged to change and wash themselves independently (self-care). However, improvements in self-care did not occur, which might be due to a lack of transfer to the tasks performed at home. As learning the skill of swimming requires inter-coordination of upper and lower limbs, and gross motor function has been reported repeatedly to improve after aquatic interventions (Ballaz et al., 2011; Dimitrijević et al., 2012; Thorpe et al., 2005), bilateral and upper limb coordination were expected to improve. However, the improvements in upper limb and bilateral coordination on dry land were lower than expected. The lower than expected improvement might be due to a lack of transfer to dry land, and to the specific items that are measured with the Bruininks-Oseretsky test of motor proficiency (2^{nd} edition) subscales.

The perceived motor competence scores showed the largest increase of all self-perception and global self-worth subscales, over the swimming intervention, improving in 4 of the 6 participants. However, no significant difference with the changes in the control group was present. The improvement in perceived motor competence over the swimming programme, which was retained after 20 weeks of follow-up (large effect size), is important as it mediates the relation between childhood motor skill proficiency and adolescent physical activity, fitness and participation levels (Barnett et al., 2008). The mean values at baseline indicated a positive self-concept; therefore, it was likely that only small improvements would occur.

Quality of life (QoL) did not change for any of the subscales over the swimming intervention, despite Groff et al. (2009) reporting that participation in adapted sports influences QoL. A reason for this lack of change might be the relatively high baseline levels compared to other literature, possibly due to the high Gross Motor Function Classification System levels included in the current sample. Since QoL is a composite of all aspects of functioning, impacted by a person's health condition, personal and environmental factors (McDougall et al., 2010), it might have been too optimistic to expect changes due to an intervention of only 10-weeks duration.
The subscales 'pain and hurt' and 'fatigue' did not change between or within groups, which was similar to the results of the pain intensity scales (Visual Analogue Scale and Faces Pain Scale–Revised) and the PedsQL^{TM} multidimensional fatigue scale, respectively. This implies that the constructs measured in these scales were similar and participants responded consistently.

Swimming skills improved significantly more over the 10-week swimming programme than over the control period, and the changes were retained with significance after a 20-week follow-up period, implying that the learned skill was consolidated. This eliminates the perception that learning a motor skill is too time-consuming for youth with CP (Verschuren et al., 2012) and is important with regard to the influence of motor proficiency on physical activity, fitness and participation levels (Bult et al., 2013; Burns et al., 2009; Kang et al., 2010; Okely, 1999; Wrotniak et al., 2006). The retention of the learned skill can facilitate engaging in swimming and other aquatic activities. Additionally, all youth had a high adherence towards the programme, participated in the intervention with high levels of enjoyment, and most youth continued to participate in swimming or aquatic activities after completing the programme. Moreover, no adverse events due to the programme were reported and no participants withdrew from the intervention. Perceived enjoyment is highly important for youth for engaging, adhering and committing to physical activity or sports (Buffart et al., 2009; Martin, 2006; Redmond & Parrish, 2008). The present study supports the relationships between skill-learning, enjoyment, adherence and continued participation.

Since the power of the statistical analyses was considerably low, no subgroup analyses were performed, as it would be impossible to draw conclusions for decisions on implementing or not implementing the intervention for the various subgroups (Brookes et al., 2004). However, from a descriptive perspective, the subgroup of youth with unilateral spasticity (4 out of 5, and 5 out of 5, respectively) improved with larger changes than the other subgroups for bilateral coordination and social functioning after the swimming intervention. Youth with dyskinesia (n = 2) improved with larger changes than the other subgroups for the subscale of Pain and Hurt of the PedsQL CP after the swimming intervention. No other subgroup-specific changes were present. The differences between subgroups for bilateral coordination could have been expected due to practising the inter-coordination of left and right, and upper and lower limbs during swimming, which is one of the main problems for youth with unilateral spasticity.

In summary, the present study showed that motor skills (walking distance at maximum walking speed, upper limb coordination, functional independence in mobility, and swimming skills) as well as perceived motor competence tended to improve after a 10-week swimming programme in youth with CP aged 7 to 17 years, with the ability to walk. These factors affect the vicious cycle of deconditioning, since motor proficiency is positively associated with levels of fitness (Burns et al., 2009), activity (Wrotniak et al., 2006) and participation (Bult et al., 2013; Kang et al., 2010; Okely, 1999) and this increase is positively mediated by perceived sports competence (Barnett et al., 2008). Based on the research of Kang et al. (2010) and Okely (1999), which found that physical functioning, sports competence, and motor proficiency improved participation, and based on the research of Buffart et al. (2009) and Redmond and Parrish (2008), which indicated that enjoyment and motivation facilitate engaging and adhering to physical activity, it was expected that the consolidation of swimming skills and enhanced motor proficiency in addition to high levels of enjoyment in the present study, would improve participation, engagement and adherence to physical activity. The present study supported these relationships. After completing the swimming programme, which was perceived as enjoyable and completed with a high attendance rate, and which improved motor skills in youth with CP, the youth participated more in swimming programmes and aquatic activities than before the start of the study.

The evidence of the quasi-experimental study suggests that participation diversity and intensity in the active-physical activities improved slightly, and the changes represented medium effect sizes. Since these activities are mostly community-based, with friends rather than family, as well as physically active (Imms et al., 2008; Shikako-Thomas et al., 2013), the improvement in diversity and intensity in these activities is important. Moreover, youth participated more with friends and others in activities of the formal domain (large effect size) and in skill-based activities (medium effect size) than at baseline, and participation occurred more in the community for the active-physical and skill-based activities (small to medium effect sizes). Since youth with CP are known to participate in activities with family at home rather than with friends in the community (Engel-Yeger et al., 2009), the increases in participation with friends in the community are of importance for the child's development. Additionally, the participants engaged in more swimming and aquatic activities than at baseline. No control data were obtained in the present study, so only tentative conclusion can be drawn. The results are promising in view of the deterioration in participation with increasing age reported in the literature for youth with CP (Law et al., 2006; Shikako-Thomas et al., 2013) The findings suggest that learning the skill of swimming encouraged participation mainly in activities of the formal domain, active-physical and skill-based activities, and facilitated youth to engage in aquatic activities, confirming the research of Kang et al. (2010), which indicated that youth who scored higher on sports and physical function, participated in more activities with friends.

The improvement in participation in formal activities with friends, in addition to the improvement in functional independence in social functioning, suggests that swimming enables youth with CP to engage in the community more independently, and with friends rather than with family.

In conclusion, the swimming skills were learned quickly and were consolidated, youth perceived the exercise as enjoyable, and engaging in the swimming programme enabled the youth to continue participating in aquatic activities. Positive trends for improvements in walking ability, upper limb coordination, functional independence in mobility and social functioning, and perceived motor competence, as well as an improvement in participation in activities of the formal domain, active-physical and skill-based activities were present after a 10-week swimming intervention in 7 to 17 year-old youth with CP with the ability to walk. Notably, the intervention was not associated with an increase in pain and fatigue, in contrast with physical therapy programmes. Swimming should be proposed to youth with CP to combat the cycle of deconditioning, as engagement and adherence were facilitated, and multiple facets of functioning at various levels of the international classification of functioning, disability and health framework were affected positively.

11.2 Practical implications

All youth had a high adherence to the swimming programme, reported high levels of enjoyment and no adverse effects were found. Swimming skills improved in all participants after the 10-week programme and the motor skills on dry land, and social and emotional well-being showed trends towards improvements.

The barriers confronting youth with CP to engaging in physical activity include the perception that learning a new skill is too time consuming, and that physical activity is not fun and carries a risk of injury, pain and fatigue (Verschuren et al., 2012). The present study showed that none of these perceptions was present for the swimming program, since in addition to high enjoyment during the programme, levels of fatigue or pain did not increase due to the swimming programme, and swimming skills were learned. These factors are important with regard to the sustainment of a physically active lifestyle. Additionally, while participating in this physical activity or sports programme, motor proficiency on land and in the water improved, as well as perceived motor competence. Therefore, an improvement in levels of physical activity, fitness and participation can be expected. Furthermore, social functioning in the daily living improved, and in activities of the formal domain participation occurred with friends and others rather than with family or alone. Finally, participation in active-physical activities, as well as in swimming programmes or aquatic activities was more frequent than at baseline. Swimming is therefore a recommended physical activity for children and adolescents with CP (with the ability to walk) to combat the cycle of deconditioning and should be promoted. Since this was the first study investigating these factors using a randomised controlled trial, and only a low statistical power of the tests was achieved, further research is necessary.

The present study showed that the inclusion of swimming in the rehabilitation programme of youth with CP may aid in breaking the cycle of deconditioning, by preventing or addressing the multiple problems that youth with CP encounter, and while youth engage in physical activity. By implementing a swimming programme within the rehabilitation programme, youth with CP might show better adherence, since the levels of enjoyment of this programme were very high, and the youth participated in aquatic activities more frequently after completing the programme, than before.

Specialists, physical therapists and parents should become aware of the benefits of a swimming programme, as it could complement part of the intensive rehabilitation programme in order to increase adherence to physical activity in adolescence. Physical activity and sport programmes should be promoted to youth with CP with trial and introductory sessions provided in community-based settings in collaboration with physical therapists, as a lack of information sustains the perception that physical activity and sport programmes are not fun and increase pain and fatigue.

11.3 Limitations of the study

The present study had a number of limitations. First, it should be kept in mind that the present study focused on children and adolescents with CP with the ability to walk, with or without hand-held mobility devices. Therefore, the results apply only to children and adolescents with CP with the ability to walk and aged between 7 and 17 years. Additionally, due to the recruitment of a convenience sample, participants can be expected to be keen on swimming, which might not reflect the general CP population. Additional limitations due to recruitment bias are reported in section 11.3.1.

Secondly, the study had a number of design-related limitations. A disadvantage of the controlled cross-over design is the carry-over or interaction effect that might be present after the follow-up period. Improvements are clinically preferred to be retained over the follow-up period. However, it conflicts with the statistical preference of absence of the carry-over effect. This conflict was dealt with in this thesis by not pooling the data. Additionally, swimming was compared to no extra exercise in the control group. Therefore, any changes that occurred might have been due to the extra time spent exercising, interacting with peers and others, and not specifically to swimming. A limitation of the quasi-experimental design investigating the effect on participation in leisure activities is, of course, the lack of a control group. Therefore it is impossible to conclude if these changes were solely due to participating in the swimming programme. The reason for not incorporating this outcome measurement in the randomised controlled cross-over design was the unlikeliness to change participation in leisure activities over a period of 10 weeks, in addition to the overlap with the 4-month period, which the participation measurement assesses.

Thirdly, the number of participants required for the present study to have sufficient statistical power (.80) was determined a priori; however this number was not reached. Therefore the power of the statistical analyses was calculated after the study was conducted, as the clinical value may be greater than the statistical significance suggests when the power is low (Portney & Watkins, 2000, p. 404). For the results of the randomised controlled cross-over design investigating the effects of the swimming intervention on pain, fatigue, walking, functional independence, coordination, self-perception, quality of life, and swimming skills, the statistical power was lower than .27. This statistical power means that the likelihood of confirming a true experimental hypothesis was in these cases only 27%. Thus, it is likely that false negatives are present in the results. The analyses of the participation data had a statistical power of .41, as a larger sample size was included in these analyses. Since the statistical power of the results was low, effect sizes, minimal important differences and other measures were used to shed more light on the results and make them more clinically interpretable. Given the low number of subjects, the results needed to be interpreted cautiously and in the light of other evidence. In particular, the increased possibility of type II errors due to the small sample size must be recognised.

Another limitation of the study was the invasive treatment 6 participants underwent during the 30-week and one-year study period. If these participants would have been excluded from the analysis, only 8 participants would have remained in the sample. The treatments included orthopaedic surgery and botulinum toxin A injections. The latter are applied frequently and therefore almost unavoidable in a research study (Heinen et al., 2010). Important to note is that none of these confounding interventions occurred during the main part of the study, i.e. the 15-week randomised controlled trial. Furthermore, participants renewed or changed orthotic devices, which might have led to differences in walking patterns, bilateral coordination, pain and QoL.

Additionally, some of the measurement tools did not meet the expectations. The assessors reported suspicion regarding the reliability of the participants' answers in the questionnaires; some of the participants did not seem to understand the questions, or seemed to be answering the questions randomly. This was the case for the pain intensity scales and the fatigue, self-perception and quality of life (QoL) questionnaires. Additionally, a discrepancy between the scores for the two pain intensity scales was observed in the results (discussed in section 5.2). The self-perception and QoL questionnaires have been reported to be reliable for use in children and adolescents with CP (Komdeur et al., 2001; Varni et al., 2006). The pain scales and the questionnaire assessing feelings of fatigue have been reported to be valid and reliable for use in typically developing children and adolescents (Gordijn et al., 2011; Stinson et al., 2006; Tomlinson et al., 2010), but no author reported the psychometric properties of these scales for youth with CP. The psychometric properties of the 'PedsQL™ cerebral palsy module version 3.0' have not been investigated for the Dutch translation that was used in the present study. Furthermore, the questionnaires assessing feelings of fatigue, QoL and self-perception have been validated for use in children from the age of 8 years onwards. Therefore, three children, aged 7 years, might have struggled to fully understand the questions. Moreover, the age limitations for some of the outcome measurements might not have been valid for all children, as some of the participants' mental age might have been lower than their chronological age.

A final limitation of the present study was that the assessor of swimming skills was not blinded to group allocation. Therefore, unintended, the assessor might have scored the participants of the swimming group higher than the control group for the water orientation measurement.

11.3.1 Recruitment problems

Recruitment was one of the largest limitations that affected the analyses of all research questions. Although the recruitment process covered a period of 6 months and addressed 25 paediatric physical therapists, a rehabilitation centre and a special school, only 15 participants took part in the baseline measurement. Other aquatic studies have used similar means of recruiting (rehabilitation centres, special education schools and paediatric physical therapists), and the maximum number of youth with CP recruited was 29 participants (Dimitrijević et al., 2012).

The results of the recruitment suggest that perhaps not all the eligible youth and families received information about the study. Additionally, a lack of time and transport were two main barriers reported by parents, as the intervention required a vast amount of time and commitment. These barriers have been reported in the literature (Shimmell et al., 2013). Some families that were approached by the therapists did report willingness to participate if the swimming programme was offered in another geographical area. On the other hand, the participating families did not withdraw from the research study, neither from the swimming sessions, nor the measurement occasions. This shows the motivation of the 14 participating families to engage in the programme. The recruitment could have been biased, as children and adolescents who were not interested in engaging in physical activity or exercise, as well as families who are experiencing a lack of support, time or transportation possibilities were less likely to be included in the present study. This part of the population might benefit the most from participating in the swimming intervention. One manner to counter this bias was the arrangement with the local special school to organise one of the two swimming sessions during school hours. For seven youth and their families participation was facilitated in this way.

11.4 Recommendations for future research

Further research should be conducted with a similar high quality research design as in the present study. Currently, randomised controlled studies are lacking in the literature. Additionally, these confirmatory studies should include larger samples than the present study, and when investigating the effect of swimming on participation in leisure activities a control group should be included. A smaller age range could be chosen to reduce some of the heterogeneity of the results, e.g. focussing on physical activity in adolescents only. However, bearing in mind the need to maximise the number of participants, this might not be feasible. A multi-centre study is advisable.

Additionally, the effect of a swimming programme on habitual physical activity could be investigated. Tools for monitoring the activity levels should include the observation of physical activity in the water.

Furthermore, other community-based exercise or physical activity programmes could be compared with the swimming programme to see the additional effect of water (due to the characteristics of water as explained in section 1.3) compared to the effects of an exercise programme in general. It is highly recommended to assess the level of enjoyment in any intervention programme, since it closely relates to adherence. The implementation of swimming into the physical therapy programme could be compared with the same amount of dry land therapy; especially long-term adherence should be assessed, due to possible differences in enjoyment and motivation to continue committing to the programmes.

Additionally, research should engage with specialists, physical therapists and parents to investigate their opinion regarding the use of physical activity such as swimming in the rehabilitation programme of youth with CP. These persons represent the main facilitators in promoting physical activity in this population, and their views and beliefs are thus important if aiming to create clinical changes.

Chapter 12: Conclusion

Children and adolescents with cerebral palsy encounter a complex set of issues that interfere with physical, emotional and social well-being. These issues are interrelated and associated with physical inactivity and the vicious cycle of deconditioning. A community-based swimming programme was proposed to combat the cycle of deconditioning as engagement and adherence were expected to be facilitated, and positive effects were expected on the multiple facets of functioning at various levels of the international classification of functioning, disability and health framework. Therefore, the aim of this study was to investigate the effect of swimming on pain intensity, perceptions of fatigue, walking ability, functional independence, coordination, perceived competence and global self-worth, quality of life, aquatic and swimming skills, as well as on participation in leisure activities, in youth with cerebral palsy with the ability to walk. Additionally the retention of possible gains was investigated. Furthermore, the enjoyment of the swimming sessions was assessed, since motivation and enjoyment are essential to sustain a physically active lifestyle.

No other randomised controlled trial has been conducted to investigate the effect of swimming on pain, fatigue, coordination, perceived competence and self-worth, and quality of life. Walking ability and functional independence have been assessed previously in controlled trials including only 3 to 6 year-olds. Furthermore, the improvement in swimming skills has not been assessed after a follow-up period longer than 3 weeks, and the enjoyment of the swimming intervention has not been reported.

A randomised controlled cross-over design was implemented and revealed that swimming skills improved significantly more over the 10-week swimming intervention in a sample of fourteen 7 to 17 year-olds with cerebral palsy with the ability to walk, than over the control period. These swimming skills were retained after a 20-week follow-up period. All youth had a high adherence towards the programme, participated in the intervention with high levels of enjoyment, and most youth continued to participate in swimming or aquatic activities after completing the programme. Moreover, no adverse events due to the programme were reported and no participants withdrew from the intervention.

The 10-week swimming intervention had a positive influence on walking distance at maximum walking speed, upper limb coordination, functional independence in social functioning and in mobility, and perceived motor competence. One year after the start of the study, in which all participants took part in the swimming intervention, the children and adolescents participated in activities of the formal domain and in skill-based activities more with friends and others than with family or alone. There was some evidence that participation diversity and intensity in active-physical activities improved, and that participation in active-physical and skill-based activities occurred more in community-based places than at home. Notably, the intervention was not associated with an increase in pain and fatigue, in contrast with physical therapy programmes. The present study showed that the perceptions of youth with cerebral palsy that learning a new skill is too time consuming, and that physical activity is not fun and carries a risk of injury, pain and fatigue, were eliminated in the swimming program. The consolidation of swimming skills and high levels of enjoyment during swimming, are expected to improve participation, engagement and adherence to physical activity, which was confirmed in the present study as youth participated more in swimming programmes and aquatic activities one year after baseline than before the start of the study. This is important with regard to the sustainment of a physically active lifestyle.

The small number of participants and the lack of control data for the participation assessment make it difficult to completely close the gap that is present in the literature. Physical, social and emotional well-being are likely to improve after a swimming programme. However, as this was the first study investigating these factors using a randomised controlled trial, and only a low statistical power of the tests was achieved, further research is necessary to conclude whether walking distance, upper limb coordination, perceived motor competence, functional independence in mobility and social functioning, and participation in leisure activities improve after a swimming programme. Further research is necessary to investigate the effects of swimming on perceived competence and quality of life, as well as on bilateral coordination and functional independence in self-care.

The findings imply that swimming is a valuable community-based exercise or physical activity that should be proposed to youth with cerebral palsy to combat the cycle of deconditioning, as engagement and adherence were facilitated, due to high levels of enjoyment, absence of increases of pain and fatigue, and skill-learning, and as multiple facets of functioning at various levels of the international classification of functioning, disability and health framework were affected positively.

References

Abel, M. F., & Damiano, D. L. (1996). Strategies for increasing walking speed in diplegic cerebral palsy. *Journal of Pediatric Orthopedics, 16*(6), 753-758.

Aidar, F. J., Silva, A. J., Reis, V. M., Careiro, A. L., Vianna, J. M., & Vovaes, G. S. (2007). Aquatic activities for severe cerebral palsy people and relation with the teach-learning process. *Fitness & Performance Journal, 6*(6), 377-381. doi: 10.3900/fpj.6.6.377.e

Aisen, M. L., Kerkovich, D., Mast, J., Mulroy, S., Wren, T. A., Kay, R. M., & Rethlefsen, S. A. (2011). Cerebral palsy: clinical care and neurological rehabilitation. *The Lancet Neurology, 10*(9), 844-852. doi: 10.1016/S1474-4422(11)70176-4

Ballaz, L., Plamondon, S., & Lemay, M. (2011). Group aquatic training improves gait efficiency in adolescents with cerebral palsy. *Disability and Rehabilitation, 33*(17-18), 1616-1624. doi: 10.3109/09638288.2010.541544

Bania, T., Dodd, K. J., & Taylor, N. (2011). Habitual physical activity can be increased in people with cerebral palsy: a systematic review. *Clinical Rehabilitation, 25*(4), 303-315. doi: 10.1177/0269215510383062

Barnett, L. M., Morgan, P. J., van Beurden, E., & Beard, J. R. (2008). Perceived sports competence mediates the relationship between childhood motor skill proficiency and adolescent physical activity and fitness: a longitudinal assessment. *The International Journal of Behavioral Nutrition and Physical Activity, 5*, 40. doi: 10.1186/1479-5868-5-40

Bates, A., & Hanson, N. (Eds.). (1996). *Aquatic exercise therapy*. Philadelphia, PA: WB Saunders Company.

Bax, M., Goldstein, M., Rosenbaum, P., Leviton, A., Paneth, N., Dan, B., . . . Damiano, D. (2005). Proposed definition and classification of cerebral palsy, April 2005. *Developmental Medicine & Child Neurology, 47*(8), 571-576. doi: 10.1017/S001216220500112X

Beckung, E., Hagberg, G., Uldall, P., & Cans, C. (2008). Probability of walking in children with cerebral palsy in Europe. *Pediatrics, 121*(1), e187-192. doi: 10.1542/peds.2007-0068

Bjornson, K. F., Belza, B., Kartin, D., Logsdon, R., & McLaughlin, J. F. (2007). Ambulatory physical activity performance in youth with cerebral palsy and youth who are developing typically. *Physical Therapy, 87*(3), 248-257. doi: 10.2522/ptj.20060157

Borggraefe, I., Kiwull, L., Schaefer, J. S., Koerte, I., Blaschek, A., Meyer-Heim, A., & Heinen, F. (2010). Sustainability of motor performance after robotic-assisted treadmill therapy in children: an open, non-randomized baseline-treatment study. *European Journal of Physical and Rehabilitation Medicine, 46*(2), 125-131. doi: Not Available

Bottos, M., & Gericke, C. (2003). Ambulatory capacity in cerebral palsy: prognostic criteria and consequences for intervention. *Developmental Medicine & Child Neurology, 45*(11), 786-790. doi: 10.1017/S0012162203001452

Brookes, S. T., Whitely, E., Egger, M., Smith, G. D., Mulheran, P. A., & Peters, T. J. (2004). Subgroup analyses in randomized trials: risks of subgroup-specific analyses; power and sample size for the interaction test. *Journal of Clinical Epidemiology, 57*(3), 229-236. doi: 10.1016/j.jclinepi.2003.08.009

Bruininks, R., & Bruininks, B. (2005). *Bruininks-Oseretsky test of motor proficiency* (2nd ed.). Minneapolis, MN: Pearson Assessments.

Buffart, L. M., Westendorp, T., van den Berg-Emons, R. J., Stam, H. J., & Roebroeck, M. E. (2009). Perceived barriers to and facilitators of physical activity in young adults with childhood-onset physical disabilities. *Journal of Rehabilitation Medicine, 41*(11), 881-885. doi: 10.2340/16501977-0420

Bull, F. C., Armstrong, T. P., Dixon, T., Ham, S., Neiman, A., & Pratt, M. (2004). Physical Inactivity. In M. Ezzati, A. D. Lopez, A. Rodgers & C. J. L. Murray (Eds.), Comparative quantification of health risks: global and regional burden of disease attributable to selected major risk factors. (Vol. 1). Geneva: World Health Organization. Retrieved from http://www.who.int/publications/cra/en/.

Bult, M. K., Verschuren, O., Gorter, J. W., Jongmans, M. J., Piskur, B., & Ketelaar, M. (2010). Cross-cultural validation and psychometric evaluation of the Dutch language version of the Children's Assessment of Participation and Enjoyment (CAPE) in children with and without physical disabilities. *Clinical Rehabilitation, 24*(9), 843-853. doi: 10.1177/0269215510367545

Bult, M. K., Verschuren, O., Lindeman, E., Jongmans, M. J., Westers, P., Claassen, A., & Ketelaar, M. (2013). Predicting leisure participation of school-aged children with cerebral palsy: longitudinal evidence of child, family and environmental factors. *Child: Care, Health and Development, 39*(3), 374-380. doi: 10.1111/j.1365-2214.2012.01391.x

Burns, Y. R., Danks, M., O'Callaghan, M. J., Gray, P. H., Cooper, D., Poulsen, L., & Watter, P. (2009). Motor coordination difficulties and physical fitness of extremely-low-birthweight children. *Developmental Medicine & Child Neurology, 51*(2), 136-142. doi: 10.1111/j.1469-8749.2008.03118.x

Butler, J. M., Scianni, A., & Ada, L. (2010). Effect of cardiorespiratory training on aerobic fitness and carryover to activity in children with cerebral palsy: a systematic review. *International Journal of Rehabilitation Research, 33*(2), 97-103. doi: 10.1097/MRR.0b013e328331c555

Butler, R. J., & Gasson, S. L. (2005). Self esteem/self concept scales for children and adolescents: a review. *Child and Adolescent Mental Health, 10*(4), 190-201. doi: 10.1111/j.1475-3588.2005.00368.x

Carlon, S. L., Taylor, N. F., Dodd, K. J., & Shields, N. (2013). Differences in habitual physical activity levels of young people with cerebral palsy and their typically developing peers: a systematic review. *Disability and Rehabilitation, 35*(8), 647-655. doi: 10.3109/09638288.2012.715721

Centre for Reviews and Dissemination, U. o. Y. (2008). Chapter 1. Core principles and methods for conducting a systematic review of health interventions.Centre for Reviews and Dissemination's Guidance for undertaking reviews in health care. University of York. Retrieved from http://www.york.ac.uk/inst/crd/pdf/Systematic_Reviews.pdf.

Chrysagis, D. N., Douka, A., Nikopoulos, M., Apostolopoulou, F., & Koutsouki, D. (2009). Effects of an aquatic program on gross motor function of children with spastic cerebral palsy. *Biology of Exercise, 5*(2), 13-25. doi: 10.4127/jbe.2009.0027

Chrysagis, N., Skordilis, E. K., Stavrou, N., Grammatopoulou, E., & Koutsouki, D. (2012). The effect of treadmill training on gross motor function and walking speed in ambulatory adolescents with cerebral palsy: a randomized controlled trial. *American Journal of Physical Medicine & Rehabilitation, 91*(9), 747-760. doi: 10.1097/PHM.0b013e3182643eba

Cohen, J. (1988). *Statistical power analysis for the behavioral sciences* (2nd ed.). Hillsdale, NJ: Lawrence Erlbaum Associates.

Cole, A., & Becker, B. (Eds.). (2004). *Comprehensive aquatic therapy* (2nd ed.). Philadelphia, PA: Butterworth-Heinemann Medical.

Coolican, H. (2009). *Research methods and statistics in psychology*. London, United Kingdom: Hodder.

Custers, J. W., van der Net, J., Hoijtink, H., Wassenberg-Severijnen, J. E., Vermeer, A., & Helders, P. J. (2002). Discriminative validity of the Dutch Pediatric Evaluation of Disability Inventory. *Archives of Physical Medicine and Rehabilitation, 83*(10), 1437-1441. doi: 10.1053/apmr.2002.34831

Custers, J. W., Wassenberg-Severijnen, J. E., Van der Net, J., Vermeer, A., Hart, H. T., & Helders, P. J. (2002). Dutch adaptation and content validity of the 'Pediatric Evaluation Of Disability Inventory (PEDI)'. *Disability and Rehabilitation, 24*(5), 250-258. doi: 10.1080/09638280110076036

Dallmeijer, A. J., & Brehm, M. A. (2011). Physical strain of comfortable walking in children with mild cerebral palsy. *Disability and Rehabilitation, 33*(15-16), 1351-1357. doi: DOI: 10.3109/09638288.2010.531374

Damiano, D. L. (2006). Activity, activity, activity: rethinking our physical therapy approach to cerebral palsy. *Physical Therapy, 86*(11), 1534-1540. doi: Not Available

de Morton, N. A. (2009). The PEDro scale is a valid measure of the methodological quality of clinical tirals: a demographic study. *Australian Journal of Physiotherapy, 55*, 129-133. doi: 10.1016/S0004-9514(09)70043-1

Deitz, J. C., Kartin, D., & Kopp, K. (2007). Review of the Bruininks-Oseretsky test of motor proficiency, 2nd edition (BOT-2). *Physical & Occupational Therapy in Pediatrics, 27*(4), 87-102. doi: 10.1080/J006v27n04_06

Demuth, S. K., Knutson, L. M., & Fowler, E. G. (2012). The PEDALS stationary cycling intervention and health-related quality of life in children with cerebral palsy: a randomized controlled trial. *Developmental Medicine & Child Neurology, 54*(7), 654-661. doi: 10.1111/j.1469-8749.2012.04321.x

Dimitrijević, L., Aleksandrovic, M., Madic, D., Okicic, T., Radovanovic, D., & Daly, D. (2012). The effect of aquatic intervention on the gross motor function and aquatic skills in children with cerebral palsy. *Journal of Human Kinetics, 32*, 167-174. doi: 10.2478/v10078-012-0033-5

Doig, G. S., & Simpson, F. (2005). Randomization and allocation concealment: a practical guide for researchers. *Journal of Critical Care, 20*(2), 187-191; discussion 191-183. doi: 10.1016/j.jcrc.2005.04.005

Du, R. Y., McGrath, C., Yiu, C. K., & King, N. M. (2010). Health- and oral health-related quality of life among preschool children with cerebral palsy. *Quality of Life Research, 19*(9), 1367-1371. doi: 10.1007/s11136-010-9693-6

Durstine, J. L., Painter, P., Franklin, B. A., Morgan, D., Pitetti, K. H., & Roberts, S. O. (2000). Physical activity for the chronically ill and disabled. *Sports Medicine, 30*(3), 207-219. doi: 10.2165/00007256-200030030-00005

Ekeland, E., Heian, F., & Hagen, K. B. (2005). Can exercise improve self esteem in children and young people? A systematic review of randomised controlled trials. *British Journal of Sports Medicine, 39*(11), 792-798. doi: 10.1136/bjsm.2004.017707

Eliasson, A. C., Krumlinde-Sundholm, L., Rosblad, B., Beckung, E., Arner, M., Ohrvall, A. M., & Rosenbaum, P. (2006). The Manual Ability Classification System (MACS) for children with cerebral palsy: scale development and evidence of validity and reliability. *Developmental Medicine & Child Neurology, 48*(7), 549-554. doi: 10.1017/S0012162206001162

Engel-Yeger, B., Jarus, T., Anaby, D., & Law, M. (2009). Differences in patterns of participation between youths with cerebral palsy and typically developing peers. *American Journal of Occupational Therapy, 63*(1), 96-104. doi: 10.5014/ajot.63.1.96

Engsberg, J. R., Ross, S. A., & Collins, D. R. (2006). Increasing ankle strength to improve gait and function in children with cerebral palsy: a pilot study. *Pediatric Physical Therapy, 18*(4), 266-275. doi: 10.1097/01.pep.0000233023.33383.2b

Epps, H., Ginnelly, L., Utley, M., Southwood, T., Gallivan, S., Scupher, M., & Woo, P. (2005). Is hydrotherapy cost-effective? A randomized controlled trial of combined hydrotherapy programmes compared with physiotherapy land techniques in children with juvenile idiopathic arthritis. *Health Technology Assessment, 9*(39), iii-iv, ix-x, 1-59. doi: 10.3310/hta9390

Fauconnier, J., Dickinson, H. O., Beckung, E., Marcelli, M., McManus, V., Michelsen, S. I., . . . Colver, A. (2009). Participation in life situations of 8-12 year old children with cerebral palsy: cross sectional European study. *British Medical Journal, 338*, b1458. doi: 10.1136/bmj.b1458

Faul, F., Erdfelder, E., Lang, A. G., & Buchner, A. (2007). G*Power 3: a flexible statistical power analysis program for the social, behavioral, and biomedical sciences. *Behavior Research Methods, 39*(2), 175-191. doi: 10.3758/BF03193146

Figuers, C. C. (2005). Aquatic therapy intervention for a child diagnosed with spinal muscular atrophy. *The Journal of Aquatic Physical Therapy, 13*(1), 6-8. doi: Not Available

Fowler, E. G., Kolobe, T. H., Damiano, D. L., Thorpe, D. E., Morgan, D. W., Brunstrom, J. E., . . . Stevenson, R. D. (2007). Promotion of physical fitness and prevention of secondary conditions for children with cerebral palsy: section on pediatrics research summit proceedings. *Physical Therapy, 87*(11), 1495-1510. doi: 10.2522/ptj.20060116

Fragala-Pinkham, M. A., Dumas, H. M., Barlow, C. A., & Pasternak, A. (2009). An aquatic physical therapy program at a pediatric rehabilitation hospital: A case series. *Pediatric Physical Therapy, 21*(1), 68-78. doi: 10.1097/PEP.0b013e318196eb37

Fragala-Pinkham, M. A., Haley, S. M., & O'Neil, M. E. (2008). Group aquatic aerobic exercise for children with disabilities. *Developmental Medicine & Child Neurology, 50*(11), 822-827. doi: 10.1111/j.1469-8749.2008.03086.x

Fragala-Pinkham, M. A., O'Neil, M. E., & Haley, S. M. (2010). Summative evaluation of a pilot aquatic exercise program for children with disabilities. *Disability and Health Journal, 3*(3), 162-170. doi: 10.1016/j.dhjo.2009.11.002

Freeman, P. R. (1989). The performance of the two-stage analysis of two-treatment, two-period crossover trials. *Statistics in Medicine, 8*(12), 1421-1432. doi: Not Available

Fritz, C. O., Morris, P. E., & Richler, J. J. (2012). Effect size estimates: current use, calculations, and interpretation. *Journal of Experimental Psychology: General, 141*(1), 2-18. doi: 10.1037/a0024338

Gates, P. E., Banks, D., Johnston, T. E., Campbell, S. R., Gaughan, J. P., Ross, S. A., . . . Tucker, C. (2012). Randomized controlled trial assessing participation and quality of life in a supported speed treadmill training exercise program vs. a strengthening program for children with cerebral palsy. *Journal of Pediatric Rehabilitation Medicine, 5*(2), 75-88. doi: 10.3233/PRM-2012-0199

Getz, M., Hutzler, Y., & Vermeer, A. (2006). Effects of aquatic interventions in children with neuromotor impairments: a systematic review of the literature. *Clinical Rehabilitation, 20*(11), 927-936. doi: 10.1177/0269215506070693

Getz, M., Hutzler, Y., & Vermeer, A. (2007). The effects of aquatic intervention on perceived physical competence and social acceptance in children with Cerebral Palsy. *European Journal for Special Needs Education, 22*, 217-228. doi: 10.108010885625070126970‌5

Getz, M., Hutzler, Y., Vermeer, A., Yarom, Y., & Unnithan, V. (2012). The effect of aquatic and land-based training on metabolic cost of walking and motor performance in children with Cerebral Palsy: a pilot study. *International Scholarly Research Network - ISRN Rehabilitation*, 1-8. doi: 10.5402/2012/657979

Gjesing, G. (2000). *Promoting activity, participation and playfulness through water activities*. Paper presented at the International Halliwick Association: Rhythm and Flow.

Gordijn, S. M., Cremers, E. M., Kaspers, G. J., & Gemke, R. J. (2011). Fatigue in children: reliability and validity of the Dutch PedsQL^{TM} Multidimensional Fatigue Scale. *Quality of Life Research*. doi: 10.1007/s11136-010-9836-9

Gorter, J. W., & Currie, S. J. (2011). Aquatic exercise programs for children and adolescents with cerebral palsy: what do we know and where do we go? *International Journal of Pediatrics, 2011*, 712165. doi: 10.1155/2011/712165

Greenleaf, C. A., Petrie, T. A., & Martin, S. B. (2010). Psychosocial variables associated with body composition and cardiorespiratory fitness in middle school students. *Research Quarterly for Exercise & Sport, 81*(3 Suppl), S65-74. doi: 10.1080/02701367.2010.10599695

Groff, D. G., Lundberg, N. R., & Zabriskie, R. B. (2009). Influence of adapted sport on quality of life: perceptions of athletes with cerebral palsy. *Disability and Rehabilitation, 31*(4), 318-326. doi: 10.1080/09638280801976233

Haley, S. M., Coster, W. J., Ludlow, L. H., Haltiwanger, J. T., & Andrellos, P. J. (2005). *PEDI-NL. Pediatric Evaluation of Disability Inventory.* (J. E. Wassenberg-Severijnen & J. W. H. Custers, Trans.). Amsterdam, The Netherlands: Harcourt Test Publishers.

Heinen, F., Desloovere, K., Schroeder, A. S., Berweck, S., Borggraefe, I., van Campenhout, A., . . . Molenaers, G. (2010). The updated European consensus 2009 on the use of botulinum toxin for children with cerebral palsy. *European Journal of Paediatric Neurology, 14*(1), 45-66. doi: 10.1016/j.ejpn.2009.09.005

Hicks, C. L., von Baeyer, C. L., Spafford, P. A., van Korlaar, I., & Goodenough, B. (2001). The Faces Pain Scale-Revised: toward a common metric in pediatric pain measurement. *Pain, 93*(2), 173-183. doi: 10.1016/S0304-3959(01)00314-1

Himpens, E., Van den Broeck, C., Oostra, A., Calders, P., & Vanhaesebrouck, P. (2008). Prevalence, type, distribution, and severity of cerebral palsy in relation to gestational age: a meta-analytic review. *Developmental Medicine & Child Neurology, 50*(5), 334-340. doi: 10.1111/j.1469-8749.2008.02047.x

Hopkins, W. G. (2000). A new view of statistics. Retrieved from http://www.sportsci.org/resource/stats/

Hopkins, W. G. (2003). A spreadsheet for analysis of straightforward controlled trials. Retrieved from http://sportsci.org/jour/03/wghtrials.htm

Hutzler, Y., Chacham, A., Bergman, U., & Reches, I. (1998). Effects of a movement and swimming program on water orientation skills and self-concept of kindergarten children with cerebral palsy. *Perceptual & Motor Skills, 86*(1), 111-118. doi: Not Available

Hutzler, Y., Chacham, A., Bergman, U., & Szeinberg, A. (1998). Effects of a movement and swimming program on vital capacity and water orientation skills of children with cerebral palsy. *Developmental Medicine & Child Neurology, 40*(3), 176-181. doi: Not Available

Imms, C., Reilly, S., Carlin, J., & Dodd, K. (2008). Diversity of participation in children with cerebral palsy. *Developmental Medicine & Child neurology, 50*(5), 363-369. doi: 10.1111/j.1469-8749.2008.02051.x

International Halliwick Association Education and Research Committee. (2010). The Halliwick Concept 2010. Retrieved from http://www.halliwick.org.uk/wp-content/uploads/2012/05/Halliwick-Concept-for-Halliwick-AST-website.pdf

Irion, J. M. (2009). Aquatic properties and therapeutic interventions. In L. T. Brody & P. R. Geigle (Eds.), *Aquatic exercise for rehabilitation and training*. Champaign, IL: Human Kinetics.

Jahnsen, R., Villien, L., Aamodt, G., Stanghelle, J. K., & Holm, I. (2003). Physiotherapy and physical activity – experiences of adults with cerebral palsy, with implications for children. *Advances in Physiotherapy, 5*, 21-32. doi: 10.1080/14038190310005779

Jahnsen, R., Villien, L., Egeland, T., Stanghelle, J. K., & Holm, I. (2004). Locomotion skills in adults with cerebral palsy. *Clinical Rehabilitation, 18*(3), 309-316. doi: 10.1191/0269215504cr735oa

Jahnsen, R., Villien, L., Stanghelle, J. K., & Holm, I. (2003). Fatigue in adults with cerebral palsy in Norway compared with the general population.

Developmental Medicine & Child Neurology, 45(5), 296-303. doi: 10.1017/S0012162203000562

Jemta, L., Fugl-Meyer, K. S., Oberg, K., & Dahl, M. (2009). Self-esteem in children and adolescents with mobility impairment: impact on well-being and coping strategies. *Acta Paediatrica, 98*(3), 567-572. doi: 10.1111/j.1651-2227.2008.01081.x

Jorgić, B., Dimitrijević, L., Aleksandrović, M., Okicić, T., Madić, D., & Radovanović, D. (2012). The swimming program effects on the gross motor function, mental adjustment to the aquatic environment, and swimming skills in children with cerebral palsy: a pilot study. *Specijalna edukacija i rehabilitacija, 11*(1), 51-66. doi: Not Available

Jorgić, B., Dimitrijević, L., Lambeck, J., Aleksandrović, M., Okicić, T., & Madić, D. (2012). Effects of aquatic programs in children and adolescents with cerebral palsy: systematic review. *Sport Science, 5*(2), 49-56. doi: Not Available

Kang, L. J., Palisano, R. J., Orlin, M. N., Chiarello, L. A., King, G. A., & Polansky, M. (2010). Determinants of social participation--with friends and others who are not family members--for youths with cerebral palsy. *Physical Therapy, 90*(12), 1743-1757. doi: 10.2522/ptj.20100048

Kelly, M., & Darrah, J. (2005). Aquatic exercise for children with cerebral palsy. *Developmental Medicine & Child neurology, 47*(12), 838-842. doi: 10.1017/S0012162205001775

Kelly, M. E., Darrah, J., Sobsey, R., & Legg, D. (2009). Effects of a community-based aquatic exercise program for children with cerebral palsy: a single subject design. *The Journal of Aquatic Physical Therapy, 17*(2), 1-11. doi: Not Available

Kerr, C., McDowell, B. C., Parkes, J., Stevenson, M., & Cosgrove, A. P. (2011). Age-related changes in energy efficiency of gait, activity, and participation in children with cerebral palsy. *Developmental Medicine & Child Neurology, 53*(1), 61-67. doi: 10.1111/j.1469-8749.2010.03795.x

King, G., Law, M., King, S., Hurley, P., Rosenbaum, P., Hanna, S., . . . Young, N. (2010). *Children's Assessment of Participation and Enjoyment and Preferences for Activities of Children. Nederlandstalige editie. Handleiding* (M. Bult & M. Ketelaar, Trans.). Amsterdam, The Netherlands: Pearson Assessment and Information.

King, G., Law, M., King, S., Rosenbaum, P., Kertoy, M. K., & Young, N. L. (2003). A conceptual model of the factors affecting the recreation and leisure participation of children with disabilities. *Physical & Occupational Therapy in Pediatrics, 23*(1), 63-90. doi: 10.1080/J006v23n01_05

Kluger, B. M., Krupp, L. B., & Enoka, R. M. (2013). Fatigue and fatigability in neurologic illnesses: proposal for a unified taxonomy. *Neurology, 80*(4), 409-416. doi: 10.1212/WNL.0b013e31827f07be

Komdeur, S. J., Schuur, R., Wijnroks, L., & Vermeer, A. (2001). De competentiebelevingsschaal voor kinderen met cerebrale parese. *Tijdschrift voor Orthopedagogiek, 40*, 579-594. doi: Not Available

Koop, S. E. (2009). Musculoskeletal growth and development. In J. R. Gage, M. H. Schwarts, S. E. Koop & T. F. Novacheck (Eds.), *The identification and*

treatment of gait problems in cerebral palsy. (2nd ed.). London: Mac Keith Press.
Lam, T., Noonan, V. K., Eng, J. J., & Team, S. R. (2008). A systematic review of functional ambulation outcome measures in spinal cord injury. *Spinal Cord, 46*(4), 246-254. doi: 10.1038/sj.sc.3102134
Lamb, K. L., Parfitt, G., & Eston, R. G. (2008). Effort perception. In N. Armstrong & W. van Mechelen (Eds.), *Pediatric Exercise Science and Medicine* (2nd ed.). Oxford, United Kingdom: Oxford University Press.
Law, M., King, G., King, S., Kertoy, M., Hurley, P., Rosenbaum, P., . . . Hanna, S. (2006). Patterns of participation in recreational and leisure activities among children with complex physical disabilities. *Developmental Medicine & Child Neurology, 48*(5), 337-342. doi: 10.1017/S0012162206000740
Levitt, S. (2010) *Treatment of cerebral palsy and motor delay.* (5th ed.). United Kingdom: Wiley-Blackwell.
Maanum, G., Jahnsen, R., Froslie, K. F., Larsen, K. L., & Keller, A. (2010). Walking ability and predictors of performance on the 6-minute walk test in adults with spastic cerebral palsy. *Developmental Medicine & Child neurology, 52*(6), e126-132. doi: 10.1111/j.1469-8749.2010.03614.x
Mackinnon, K. (1997). An evaluation of the benefits of Halliwick swimming on a child with mild spastic diplegia. *Association of Paediatric Chartered Physiotherapists Journal*, 30-39. doi: Not Available
Maher, C. A., Olds, T., Williams, M. T., & Lane, A. E. (2008). Self-reported quality of life in adolescents with cerebral palsy. *Physical & Occupational Therapy in Pediatrics, 28*(1), 41-57. doi: 10.1300/J006v28n01_04
Majnemer, A., Shevell, M., Law, M., Birnbaum, R., Chilingaryan, G., Rosenbaum, P., & Poulin, C. (2008). Participation and enjoyment of leisure activities in school-aged children with cerebral palsy. *Developmental Medicine & Child Neurology, 50*(10), 751-758. doi: 10.1111/j.1469-8749.2008.03068.x
Maltais, D. B., Dumas, F., Boucher, N., & Richards, C. L. (2010). Factors related to physical activity in adults with cerebral palsy may differ for walkers and nonwalkers. *American Journal of Physical Medicine & Rehabilitation, 89*(7), 584-597. doi: 10.1097/PHM.0b013e3181d89f32
Martin, J. J. (2006). Psychosocial aspects of youth disability sport. *Adapted Physical Activity Quarterly, 23*(1), 65-77. doi: Not Available
McDougall, J., Wright, V., & Rosenbaum, P. (2010). The ICF model of functioning and disability: incorporating quality of life and human development. *Developmental Neurorehabilitation, 13*(3), 204-211. doi: 10.3109/17518421003620525
McDowell, B. C., Humphreys, L., Kerr, C., & Stevenson, M. (2009). Test-retest reliability of a 1-minute walk test in children with bilateral spastic cerebral palsy. *Gait Posture, 29*(2), 267-269. doi: 10.1016/j.gaitpost.2008.09.010
McDowell, B. C., Kerr, C., Parkes, J., & Cosgrove, A. (2005). Validity of a 1 minute walk test for children with cerebral palsy. *Developmental Medicine & Child Neurology, 47*(11), 744-748. doi: 10.1017/S0012162205001568
McManus, B. M., & Kotelchuck, M. (2007). The effect of aquatic therapy on functional mobility of infants and toddlers in early intervention. *Pediatric Physical Therapy, 19*(4), 275-282. doi: 10.1097/PEP.0b013e3181575190

Mehta, C. R., & Patel, N. R. (1989, 2010). *IBM SPSS Exact tests.* Cambridge, Massachusetts: Cytel Software Corporation and Harvard School of Public Health.

Mesterman, R., Leitner, Y., Yifat, R., Gilutz, G., Levi-Hakeini, O., Bitchonsky, O., . . . Harel, S. (2010). Cerebral palsy--long-term medical, functional, educational, and psychosocial outcomes. *Journal of Child Neurology, 25*(1), 36-42. doi: 10.1177/0883073809336677

Miller, F. (2005). Gait. *Cerebral Palsy*. New York: Springer Science.

Morgan, P., & McGinley, J. (2013). Gait function and decline in adults with cerebral palsy: a systematic review. *Disability and Rehabilitation*, advanced online publication. doi: 10.3109/09638288.2013.775359

Munro, B. H. (2005). *Statistical methods for health care research* (5^{th} ed.). Philadelphia, PA: Lippincott Williams & Wilkins.

Murphy, N. A., & Carbone, P. S. (2008). Promoting the participation of children with disabilities in sports, recreation, and physical activities. *Pediatrics, 121*(5), 1057-1061. doi: 10.1542/peds.2008-0566

National Institute for Health and Clinical Excellence. (2006). *Methods for development of NICE public health guidance*

Nieuwenhuijsen, C., Donkervoort, M., Nieuwstraten, W., Stam, H. J., & Roebroeck, M. E. (2009). Experienced problems of young adults with cerebral palsy: targets for rehabilitation care. *Archives of Physical Medicine and Rehabilitation, 90*(11), 1891-1897. doi: 10.1016/j.apmr.2009.06.014

Ohata, K., Tsuboyama, T., Haruta, T., Ichihashi, N., Kato, T., & Nakamura, T. (2008). Relation between muscle thickness, spasticity, and activity limitations in children and adolescents with cerebral palsy. *Developmental Medicine & Child Neurology, 50*(2), 152-156. doi: 10.1111/j.1469-8749.2007.02018.x

Öhrvall, A. M., Eliasson, A. C., Löwing, K., Ödman, P., & Krumlinde-Sundholm, L. (2010). Self-care and mobility skills in children with cerebral palsy, related to their manual ability and gross motor function classifications. *Developmental Medicine & Child Neurology, 52*(11), 1048-1055. doi: 10.1111/j.1469-8749.2010.03764.x

Okely, A. D. (1999). *The relationship of participation in organised sports and games, participation in nonorganised physical activity, and cardiorespiratory endurance to fundamental motor skill ability among adolescents.* University of Wollongong.

Oskoui, M., Coutinho, F., Dykeman, J., Jetté, N., & Pringsheim, T. (2013). An update on the prevalence of cerebral palsy: a systematic review and meta-analysis. *Developmental Medicine & Child Neurology, 55*(6), 509-519. doi: 10.1111/dmcn.12080

Őzer, D., Nalbant, S., Aktop, A., Duman, O., Keles, I., & Toraman, N. F. (2007). Swimming training program for children with cerebral palsy: body perceptions, problem behaviour, and competence. *Perceptual and Motor Skills, 105*(3 Pt 1), 777-787. doi: 10.2466/pms.105.3.777-787

Palisano, R. J., Kang, L. J., Chiarello, L. A., Orlin, M., Oeffinger, D., & Maggs, J. (2009). Social and community participation of children and youth with cerebral palsy is associated with age and gross motor function classification. *Physical Therapy, 89*(12), 1304-1314. doi: 10.2522/ptj.20090162

Palisano, R. J., Rosenbaum, P., Walter, S., Russell, D., Wood, E., & Galuppi, B. (1997). Development and reliability of a system to classify gross motor function in children with cerebral palsy. *Developmental Medicine & Child Neurology, 39*(4), 214-223. doi: 10.1111/j.1469-8749.1997.tb07414.x

Parkinson, K. N., Dickinson, H. O., Arnaud, C., Lyons, A., Colver, A., & group, S. (2013). Pain in young people aged 13 to 17 years with cerebral palsy: cross-sectional, multicentre European study. *Archives of Disease in Childhood, 98*(6), 434-440. doi: 10.1136/archdischild-2012-303482

Parkinson, K. N., Gibson, L., Dickinson, H. O., & Colver, A. F. (2010). Pain in children with cerebral palsy: a cross-sectional multicentre European study. *Acta Paediatrica, 99*(3), 446-451. doi: 10.1111/j.1651-2227.2009.01626.x

Penner, M., Xie, W. Y., Binepal, N., Switzer, L., & Fehlings, D. (2013). Characteristics of pain in children and youth with cerebral palsy. *Pediatrics*. doi: 10.1542/peds.2013-0224

Pett, M. A. (1997). *Nonparametric statistics for health care research: statistics for small samples and unusual distributions*. California: SAGE Publications.

Pfeiffer, K. A., Pivarnik, J. M., Womack, C. J., Reeves, M. J., & Malina, R. M. (2002). Reliability and validity of the Borg and OMNI rating of perceived exertion scales in adolescent girls. *Medicine & Science in Sports & Exercise, 34*(12), 2057-2061. doi: 10.1249/01.MSS.0000039302.54267.BF

Portney, L. G., & Watkins, M. P. (2000). *Foundations of Clinical Research. Applications to practice* (2nd ed.). New Jersey: Prentice Hall Health.

Ramstad, K., Jahnsen, R., Skjeldal, O. H., & Diseth, T. H. (2011). Characteristics of recurrent musculoskeletal pain in children with cerebral palsy aged 8 to 18 years. *Developmental Medicine & Child Neurology, 53*(11), 1013-1018. doi: 10.1111/j.1469-8749.2011.04070.x

Ramstad, K., Jahnsen, R., Skjeldal, O. H., & Diseth, T. H. (2012). Parent-reported participation in children with cerebral palsy: the contribution of recurrent musculoskeletal pain and child mental health problems. *Developmental Medicine & Child Neurology, 54*(9), 829-835. doi: 10.1111/j.1469-8749.2012.04341.x

Ratkowsky, D. A., Evans, M. A., & Alldredge, J. R. (1993). *Cross-over experiments: design, analysis, and application*. New York: Marcel Decker.

Redman, T. A., Finn, J. C., Bremner, A. P., & Valentine, J. (2008). Effect of upper limb botulinum toxin-A therapy on health-related quality of life in children with hemiplegic cerebral palsy. *Journal of Paediatrics and Child Health, 44*(7-8), 409-414. doi: 10.1111/j.1440-1754.2008.01319.x

Redmond, R., & Parrish, M. (2008). Variables influencing physiotherapy adherence among young adults with cerebral palsy. *Qualitative Health Research, 18*(11), 1501-1510. doi: 10.1177/1049732308325538

Retarekar, R., Fragala-Pinkham, M. A., & Townsend, E. L. (2009). Effects of aquatic aerobic exercise for a child with cerebral palsy: single-subject design. *Pediatric Physical Therapy, 21*(4), 336-344. doi: 10.1097/PEP.0b013e3181beb039

Riad, J., Brostrom, E., & Langius-Eklof, A. (2013). Do movement deviations influence self-esteem and sense of coherence in mild unilateral cerebral palsy? *Journal of Pediatric Orthopaedics, 33*(3), 298-302. doi: 10.1097/BPO.0b013e31827d0b5b

Rimmer, J. H. (1999). Health promotion for people with disabilities: the emerging paradigm shift from disability prevention to prevention of secondary conditions. *Physical Therapy, 79*(5), 495-502.

Rimmer, J. H. (2001). Physical fitness levels of persons with cerebral palsy. *Developmental Medicine & Child Neurology, 43*(3), 208-212. doi: 10.1111/j.1469-8749.2001.tb00189.x

Rimmer, J. H. (2005). Exercise and physical activity in persons aging with a physical disability. *Physical Medicine & Rehabilitation Clinics of North America, 16*(1), 41-56. doi: 10.1016/j.pmr.2004.06.013

Riner, W. F., & Sellhorst, S. H. (2013). Physical activity and exercise in children with chronic health conditions. *Journal of Sport and Health Science, 2*, 12-20. doi: 10.1016/j.jshs.2012.11.005

Robertson, R. J., Goss, F. L., Boer, N. F., Peoples, J. A., Foreman, A. J., Dabayebeh, I. M., . . . Thompkins, T. (2000). Children's OMNI scale of perceived exertion: mixed gender and race validation. *Medicine & Science in Sports & Exercise, 32*(2), 452-458.

Rogers, A., Furler, B. L., Brinks, S., & Darrah, J. (2008). A systematic review of the effectiveness of aerobic exercise interventions for children with cerebral palsy: an AACPDM evidence report. *Developmental Medicine & Child Neurology, 50*(11), 808-814. doi: 10.1111/j.1469-8749.2008.03134.x

Rosenbaum, P., Paneth, N., Leviton, A., Goldstein, M., Bax, M., Damiano, D., . . . Jacobsson, B. (2007). A report: the definition and classification of cerebral palsy April 2006. *Developmental Medicine & Child Neurology. Supplement, 109*, 8-14. doi: 10.1111/j.1469-8749.2007.tb12610.x

Rossier, P., & Wade, D. T. (2001). Validity and reliability comparison of 4 mobility measures in patients presenting with neurologic impairment. *Archives of Physical Medicine and Rehabilitation, 82*(1), 9-13. doi: 10.1053/apmr.2001.9396

Ruskin, D. A., Amaria, K. A., Warnock, F. F., & McGrath, P. A. (2011). Assessment of pain in infants, children and adolescents. In D. C. Turk & R. Melzack (Eds.), *Handbook of pain assessment.* (3rd ed.). New York: The Guilford Press.

Russo, R. N., Goodwin, E. J., Miller, M. D., Haan, E. A., Connell, T. M., & Crotty, M. (2008). Self-esteem, self-concept, and quality of life in children with hemiplegic cerebral palsy. *Journal of Pediatrics, 153*(4), 473-477. doi: 10.1016/j.jpeds.2008.05.040

Russo, R. N., Miller, M. D., Haan, E., Cameron, I. D., & Crotty, M. (2008). Pain characteristics and their association with quality of life and self-concept in children with hemiplegic cerebral palsy identified from a population register. *The Clinical Journal of Pain, 24*(4), 335-342. doi: 10.1097/AJP.0b013e318162eae0

Sackett, D. L. (1989). Rules of evidence and clinical recommendations on the use of antithrombotic agents. *Chest, 95*(2 Suppl), 2S-4S. doi: Not Available

Salem, Y., & Gropack, S. J. (2010). Aquatic therapy for a child with type III spinal muscular atrophy: a case report. *Physical & Occupational Therapy in Pediatrics, 30*(4), 313-324. doi: 10.3109/01942638.2010.493097

Scholtes, V. A., Becher, J. G., Janssen-Potten, Y. J., Dekkers, H., Smallenbroek, L., & Dallmeijer, A. J. (2012). Effectiveness of functional progressive resistance exercise training on walking ability in children with cerebral palsy: a randomized controlled trial. *Research in Developmental Disabilities, 33*(1), 181-188. doi: 10.1016/j.ridd.2011.08.026

Schuengel, C., Voorman, J., Stolk, J., Dallmeijer, A., Vermeer, A., & Becher, J. (2006). Self-worth, perceived competence, and behaviour problems in children with cerebral palsy. *Disability and Rehabilitation, 28*(20), 1251-1258. doi: 10.1080/09638280600554652

Schulz, K. F., Altman, D. G., & Moher, D. (2010). CONSORT 2010 Statement: updated guidelines for reporting parallel group randomised trials. *BMC Medicine, 8*, 18. doi: 10.1186/1741-7015-8-18

Scottish Intercollegiate Guidelines Network. (2008). SIGN 50: A guideline developer's handbook. Algorithm for classifying study design for questions of effectiveness. Retrieved from http://www.sign.ac.uk/pdf/studydesign.pdf

Sellier, E., Surman, G., Himmelmann, K., Andersen, G., Colver, A., Krageloh-Mann, I., . . . Cans, C. (2010). Trends in prevalence of cerebral palsy in children born with a birthweight of 2,500 g or over in Europe from 1980 to 1998. *European Journal of Epidemiology, 25*(9), 635-642. doi: 10.1007/s10654-010-9474-0

Senn, S. (1993). *Cross-over trials in clinical research*. Chichester: Wiley.

Senn, S. (2006). Cross-over trials in Statistics in Medicine: the first '25' years. *Statistics in Medicine, 25*(20), 3430-3442. doi: 10.1002/sim.2706

Shen, D. (2006). *Estimate carryover effect in clinical trial crossover designs*. Paper presented at the Proceedings SAS Conference, PharmaSUG, Bonita Springs, FL.

Shields, N., Loy, Y., Murdoch, A., Taylor, N. F., & Dodd, K. J. (2007). Self-concept of children with cerebral palsy compared with that of children without impairment. *Developmental Medicine & Child Neurology, 49*(5), 350-354. doi: 10.1111/j.1469-8749.2007.00350.x

Shikako-Thomas, K., Shevell, M., Lach, L., Law, M., Schmitz, N., Poulin, C., . . . group, Q. (2013). Picture me playing – a portrait of participation and enjoyment of leisure activities in adolescents with cerebral palsy. *Research in Developmental Disabilities, 34*(3), 1001-1010. doi: 10.1016/j.ridd.2012.11.026

Shimmell, L. J., Gorter, J. W., Jackson, D., Wright, M., & Galuppi, B. (2013). "It's the participation that motivates him": physical activity experiences of youth with cerebral palsy and their parents. *Physical & Occupational Therapy in Pediatrics*. doi: 10.3109/01942638.2013.791916

Siebes, R. C., Wijnroks, L., & Vermeer, A. (2002). Qualitative analysis of therapeutic motor intervention programmes for children with cerebral palsy: an update. *Developmental Medicine & Child Neurology, 44*(9), 593-603. doi: 10.1017/S0012162201002638

Siegel, S., & Castellan, N. J. (1988). Non parametric statistics for the behavioral sciences (pp. 180-181). Singapore: McGraw-Hill.

Soyupek, F., Aktepe, E., Savas, S., & Askin, A. (2010). Do the self-concept and quality of life decrease in CP patients? Focussing on the predictors of self-concept and quality of life. *Disability and Rehabilitation, 32*(13), 1109-1115. doi: 10.3109/09638280903391120

Stinson, J. N., Kavanagh, T., Yamada, J., Gill, N., & Stevens, B. (2006). Systematic review of the psychometric properties, interpretability and feasibility of self-report pain intensity measures for use in clinical trials in children and adolescents. *Pain, 125*(1-2), 143-157. doi: 10.1016/j.pain.2006.05.006

Surveillance of Cerebral Palsy in Europe. (2002). Prevalence and characteristics of children with cerebral palsy in Europe. *Developmental Medicine & Child neurology, 44*(9), 633-640. doi: 10.1111/j.1469-8749.2002.tb00848.x

Tantilipikorn, P., Watter, P., & Prasertsukdee, S. (2013). Feasibility, reliability and validity of the Thai version of the Pediatric Quality of Life Inventory 3.0 cerebral palsy module. *Quality of Life Research, 22*(2), 415-421. doi: 10.1007/s11136-012-0161-3

Tarsuslu, T., & Livanelioglu, A. (2010). Relationship between quality of life and functional status of young adults and adults with cerebral palsy. *Disability and Rehabilitation, 32*(20), 1658-1665. doi: 10.3109/09638281003649904

Telama, R., Yang, X., Viikari, J., Valimaki, I., Wanne, O., & Raitakari, O. (2005). Physical activity from childhood to adulthood: a 21-year tracking study. *American Journal of Preventive Medicine, 28*(3), 267-273. doi: 10.1016/j.amepre.2004.12.003

Thorpe, D., Reilly, M., & Case, L. (2005). The effects of an aquatic resistive exercise program on ambulatory children with cerebral palsy. *The Journal of Aquatic Physical Therapy 13*, 21-34. doi: Not Available

Tirosh, R., Katz-Leurer, M., & Getz, M. D. (2008). Halliwick-Based Aquatic Assessments: Reliability and Validity. *International Journal of Aquatic Research & Education, 2*(3), 224. doi: Not Available

Tomlinson, D., von Baeyer, C. L., Stinson, J. N., & Sung, L. (2010). A systematic review of faces scales for the self-report of pain intensity in children. *Pediatrics, 126*(5), e1168-1198. doi: 10.1542/peds.2010-1609

Unger, M., Jelsma, J., & Stark, C. (2013). Effect of a trunk-targeted intervention using vibration on posture and gait in children with spastic type cerebral palsy: a randomized control trial. *Developmental Neurorehabilitation, 16*(2), 79-88. doi: 10.3109/17518423.2012.715313

Utter, A. C., Robertson, R. J., Nieman, D. C., & Kang, J. (2002). Children's OMNI scale of perceived exertion: walking/running evaluation. *Medicine & Science in Sports & Exercise, 34*(1), 139-144. doi: 10.1097/00005768-200201000-00021

Van der Slot, W. M., Nieuwenhuijsen, C., Van den Berg-Emons, R. J., Bergen, M. P., Hilberink, S. R., Stam, H. J., & Roebroeck, M. E. (2012). Chronic pain, fatigue, and depressive symptoms in adults with spastic bilateral cerebral palsy. *Developmental Medicine & Child Neurology, 54*, 836-842. doi: 10.1111/j.1469-8749.2012.04371.x

Van Hedel, H. J., Wirz, M., & Dietz, V. (2005). Assessing walking ability in subjects with spinal cord injury: validity and reliability of 3 walking tests. *Archives of Physical Medicine and Rehabilitation, 86*(2), 190-196. doi: 10.1016/j.apmr.2004.02.010

Van Wely, L., Becher, J. G., Balemans, A. C. J., & Dallmeijer, A. (2012). Ambulatory activity of children with cerebral palsy: which characteristics are important. *Developmental Medicine & Child Neurology, 54*, 436-442. doi: DOI: 10.1111/j.1469-8749.2012.04251.x

Vargus-Adams, J. (2005). Health-related quality of life in childhood cerebral palsy. *Archives of Physical Medicine and Rehabilitation, 86*(5), 940-945. doi: 10.1016/j.apmr.2004.10.036

Varni, J. W., Burwinkle, T. M., Berrin, S. J., Sherman, S. A., Artavia, K., Malcarne, V. L., & Chambers, H. G. (2006). The PedsQL™ in pediatric cerebral palsy: reliability, validity, and sensitivity of the Generic Core Scales and Cerebral Palsy Module. *Developmental Medicine & Child Neurology, 48*(6), 442-449. doi: 10.1017/S001216220600096X

Varni, J. W., Burwinkle, T. M., Katz, E. R., Meeske, K., & Dickinson, P. (2002). The PedsQL™ in pediatric cancer: Reliability and validity of the Pediatric Quality of Life Inventory™ Generic Core Scales, Multidimensional Fatigue Scale, and Cancer Module. *Cancer, 94*, 2090-2106. doi: 10.1002/cncr.10428

Varni, J. W., Burwinkle, T. M., Sherman, S. A., Hanna, K., Berrin, S. J., Malcarne, V. L., & Chambers, H. G. (2005). Health-related quality of life of children and adolescents with cerebral palsy: hearing the voices of the children. *Developmental Medicine & Child Neurology, 47*(9), 592-597. doi: 10.1017/S0012162205001179

Varni, J. W., Burwinkle, T. M., & Szer, I. S. (2004). The PedsQL™ Multidimensional Fatigue Scale in pediatric rheumatology: Reliability and validity. *Journal of Rheumatology, 31*, 2494-2500. doi: Not Available

Varni, J. W., & Limbers, C. A. (2008). The PedsQL™ Multidimensional Fatigue Scale in young adults: Feasibility, reliability and validity in a university student population. *Quality of Life Research, 17*, 105-114. doi: 10.1007/s11136-007-9282-5

Veerman, J. W., Straathof, M. A. E., Treffers, P. D. A., Van den Bergh, B. R. H., & ten Brink, L. T. (1997). *Competentiebelevingsschaal voor kinderen - handleiding*. Lisse, The Netherlands: Swets & Zeitlinger B.V.

Verschuren, O., Ketelaar, M., Gorter, J. W., Helders, P. J., Uiterwaal, C. S., & Takken, T. (2007). Exercise training program in children and adolescents with cerebral palsy: a randomized controlled trial. *Archives of Pediatrics and Adolescent Medicine, 161*(11), 1075-1081. doi: 10.1001/archpedi.161.4.356

Verschuren, O., Ketelaar, M., Takken, T., Helders, P. J., & Gorter, J. W. (2008). Exercise programs for children with cerebral palsy: a systematic review of the literature. *American Journal of Physical Medicine & Rehabilitation, 87*(5), 404-417. doi: 10.1097/PHM.0b013e31815b2675

Verschuren, O., & Takken, T. (2010). Aerobic capacity in children and adolescents with cerebral palsy. *Research in Developmental Disabilities, 31*(6), 1352-1357. doi: 10.1016/j.ridd.2010.07.005

Verschuren, O., Wiart, L., Hermans, D., & Ketelaar, M. (2012). Identification of facilitators and barriers to physical activity in children and adolescents with cerebral palsy. *Journal of Pediatrics, 161*(3), 488-494. doi: 10.1016/j.jpeds.2012.02.042

Von Baeyer, C. L. (2006). Children's self-reports of pain intensity: Scale selection, limitations and interpretation. *Pain Research & Management, 11*(3), 157-162. doi: Not Available

Ward, A. B. (2008). Spasticity treatment with botulinum toxins. *Journal of Neural Transmission, 115*(4), 607-616. doi: 10.1007/s00702-007-0833-2

Wassenberg-Severijnen, J. E., Custers, J. W., Hox, J. J., Vermeer, A., & Helders, P. J. (2003). Reliability of the Dutch Pediatric Evaluation of Disability Inventory (PEDI). *Clinical Rehabilitation, 17*(4), 457-462. doi: 10.1191/0269215503cr634oa

Willoughby, K. L., Dodd, K. J., Shields, N., & Foley, S. (2010). Efficacy of partial body weight-supported treadmill training compared with overground walking practice for children with cerebral palsy: a randomized controlled trial. *Archives of Physical Medicine and Rehabilitation, 91*(3), 333-339. doi: 10.1016/j.apmr.2009.10.029

World Health Organization. (2007). *ICF: International Classification of Functioning, Disability and Health. Children and Youth version*. Geneva.

Wrotniak, B. H., Epstein, L. H., Dorn, J. M., Jones, K. E., & Kondilis, V. A. (2006). The relationship between motor proficiency and physical activity in children. *Pediatrics, 118*(6), e1758-1765. doi: 10.1542/peds.2006-0742

Wuang, Y. P., & Su, C. Y. (2009). Reliability and responsiveness of the Bruininks-Oseretsky test of motor proficiency 2nd edition in children with intellectual disability. *Research in Developmental Disabilities, 30*(5), 847-855. doi: 10.1016/j.ridd.2008.12.002

Ziviani, J., Ottenbacher, K. J., Shephard, K., Foreman, S., Astbury, W., & Ireland, P. (2002). Concurrent validity of the Functional Independence Measure for Children (WeeFIM) and the Pediatric Evaluation of Disabilities Inventory in children with developmental disabilities and acquired brain injuries. *Physical & Occupational Therapy in Pediatrics, 21*(2-3), 91-101. doi: 10.1080/J006v21n02_08

Appendices

A. Intervention details of the articles discussed in the systematic review
B. Recruitment letter and poster
C. Informed consent
D. Stratification and randomisation process
E. Order of measurements; Bruininks-Oseretksy test of motor proficiency (2nd edition) Record form; Level of perceived exertion and enjoyment rating scales
F. Decision making models
G. List of publications, including a copy of the published manuscript